

THE EFFECTS OF CHILD COMPLEXITY ON RESPONSES TO BEHAVIOURAL
INTERVENTIONS FOR SLEEP PROBLEMS IN CHILDREN WITH AUTISM SPECTRUM
DISORDERS

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Abstract

Sleep problems are one of the most commonly reported concerns among parents of children with Autism Spectrum Disorder (ASD). Unless treated effectively, sleep problems are highly persistent and sometimes continue into adulthood. Given the prevalence of sleep problems and long-term effects associated with sleep problems (i.e., impaired daytime and adaptive functioning, social interaction and communication difficulties), it is crucial that effective treatments are identified. Behavioural treatments such as scheduled awakenings and extinction meet the criteria to be considered possibly efficacious intervention and more future replications are needed in order to be considered well-established interventions. Similarly, in the extant literature, there is an emerging body of evidence demonstrating the effectiveness of behaviourally-based, parent-implemented sleep treatments that are informed by functional behavioural assessment (FBA). However, further replication and expansion of this research is also required. As a starting point, factors that may affect response to behaviourally-based, parent-implemented sleep treatments need to be investigated and identified.

The purpose of this study was, therefore, to investigate and identify the complex array of factors that may affect responses to FBA-informed and parent-implemented behavioural sleep interventions. Following Kazdin and Whitley (2006), various factors associated with the scope and severity of the child's problem were used to define the dimensions of clinical complexity. These factors included impaired daytime functioning and behaviour, sleep problems, presence of comorbid psychopathology and medications use. Impaired daytime functioning and behaviour were measured using the Child Behaviour Checklist (CBCL), the Gilliam Autism Rating Scale, Second Edition (GARS-2), and the Vineland Adaptive Behaviour Scale, Second Edition (VABS II). Children's sleep problems were measured using the Children's Sleep Habits Questionnaire (CSHQ). Presence of comorbid psychopathology was measured using a categorical variable of comorbidity and medications use was measured using a categorical variable of medications. A Sleep Problem Severity (SPS) score was calculated for each child in order to objectively measure the changes

before and after treatment. A pretest-posttest research design was used to analyse existing clinical data obtained from 31 children who had parent reported sleep disturbance, and who had received an FBA-informed intervention. Results from paired sample t test showed that there was a statistically significant reduction in sleep problem severity between pre- and post-treatment. Based on evaluation of modified Brinley plots, 27 out of the of 31 children showed improvement post-treatment. Factor analysis identified three latent variables underlying behaviour problems, sleep problems, autism severity, communications, medications and comorbidity. The latent variable *Behaviour and Sleep Problems* was identified as a statistically significant predictor of changes in SPS score post-treatment. The second latent variable *Medication-Communication* was not a statistically significant predictor of changes in SPS score post-treatment. The third latent variable *Psychopathology Severity* was also not a statistically significant predictor of changes in SPS score post-treatment. The present findings add to the limited literature investigating the child characteristics that may impact upon response to behavioural sleep interventions in children with ASD. In terms of treatment, additional components to address behavioural problems will need to be formulated and included as part of the sleep intervention treatment package so that both behaviour and sleep problems can be addressed together to enhance treatment responses. Further research is still needed to develop a deeper understanding of the complex array of factors that may affect treatment responses.

Chapter 1

Introduction

Research suggests that up to 80% of children and adolescents (referred to as children throughout the rest of this thesis) with Autism Spectrum Disorder (ASD) experience some form of sleep problem (Kose, Yilmaz, Ocakoglu, & Ozbaran, 2017; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Mannion, Leader, & Healy, 2013; Singh & Zimmerman, 2015; Turner & Johnson, 2012). This is compared to only 25% to 50% of typically developing children (Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014). The most commonly reported sleep problems in children with developmental disorders, of which ASD is a sub-set, are classified as behavioural insomnias (Souders et al., 2009). This includes sleep onset delay, refusal to sleep alone, short sleep duration, settling difficulties, frequent night wakings, early morning wakings, irregular/problematic bedtime routines, and excessive daytime sleepiness (Richdale & Schreck, 2009). Without intervention, children with ASD are unlikely to outgrow their sleep problems (Hodge et al., 2014), and in many cases, they might in fact worsen over time (Sivertsen, Posserud, Gillberg, Lundervold, & Hysing, 2012).

Sleep problems in children with ASD have been associated with increased severity of autism symptomatology, including greater communication and social skills difficulties, and restricted and repetitive behaviours (Cortesi, Giannotti, Ivanenko, & Johnson, 2010; Goldman et al., 2009; Hollway & Aman, 2011; Malow, McGrew, Harvey, Henderson, & Stone, 2006; Richdale & Schreck, 2009; Schreck, Mulick, & Smith, 2004), increased rates of internalising and externalising behaviour problems and poorer adaptive skill development (Sikora, Johnson, Clemons, & Katz, 2012). Given the prevalence of sleep problems in children with ASD, and the significant association found between sleep problems and daytime functioning, early identification and treatment is essential.

To date, systematic reviews have identified that both melatonin (Guenole & Baleyte, 2011; Hollway & Aman, 2011; Rossignol & Frye, 2011; Sung, Fung, Cai, & Ooi, 2010) and behavioural interventions (Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006; Turner & Johnson, 2012; Vriend, Corkum, Moon, & Smith, 2011) are effective in addressing sleep problems in children with ASD. However, we know very little about child variables (e.g., behaviour problems, sleep problems, medications, comorbid psychopathology) that may effect treatment outcome. The aim of the current study is to enhance our understanding of the relationship between child complexity variables and response to functional behavioural assessment (FBA) based intervention among children with ASD and sleep problems.

This chapter provides an overview of autism and associated challenging behaviours, common behavioural insomnias in children with autism, types and causes of sleep problems in children with autism, common treatments for sleep problems, and case complexity and treatment responses.

Autism Spectrum Disorder

Definition. Autism Spectrum Disorder (ASD) is a complex neurodevelopmental disorder characterised by impairments in social communication and interaction and restricted, repetitive patterns of behaviour (American Psychiatric Association [APA], 2016). Impairments in communication could include difficulties with language comprehension and expression (e.g., idiosyncratic language use, echolalia) and difficulties with nonverbal communication such as hand gestures or body language, and limited facial expressions. Impairments in social interaction could include difficulties in interacting with others, establishing and maintaining friendships, making eye contact, and recognising facial expressions. Restricted and repetitive behavioural patterns may include vocal stereotypy and repetitive body movements (e.g., body rocking or hand flapping) (APA, 2016). Autism is a largely heterogeneous disorder and the extent of impairment varies greatly across individuals (Waterhouse et al., 1996). The heterogeneity in clinical presentation is

heavily influenced by several factors such as genetic variability, comorbidity, and gender (Masi, DeMayo, Glozier, & Guastella, 2017).

Prevalence. Autism Spectrum Disorder is about three to four times more common among boys than girls (Baio et al., 2014; Lord, Elsabbagh, Baird, & Veenstra-Vanderweele, 2018). In the United States of America, it is estimated that 1 in 43 children have a diagnosis of ASD (Xu, Strathearn, Liu, & Bao, 2018). This compares to a prevalence of 1 in 63 children (Baron-Cohen et al., 2009) in the United Kingdom, and 1 in 675 children in Asia (Sun & Allison, 2010). Globally, in 2010, the population prevalence was estimated to be 1 in 132 persons (Baxter et al., 2015). Prevalence data is unavailable in New Zealand; however, it is estimated that approximately 1 in 100 people in this country have a diagnosis of ASD (Ministries of Health and Education, 2016).

Etiology. The etiology of ASD is largely unknown; however, it is thought to be the result of a combination of genetic, biological, and environmental factors.

Genetic factors. Neurobiological and genetic factors have been identified in the past decade (Lyall et al., 2017) with estimates suggesting that the heritability of autism ranges from 50-90% (Fakhoury, 2015; Sandin et al., 2014). Two recent twin studies examined concordance rates between monozygotic and dizygotic twin pairs and found that concordance for autism in monozygotic twin pairs was typically at least triple that in dizygotic twin pairs indicated that autism was predominantly caused by genetic factors (Colvert et al., 2015; Nordenbaek, Jorgensen, Ohm Kyvik, & Bilenberg, 2014). Colvert et al. (2015) examined the genetic and environmental contributions to autism in a population-based sample of twins born in England and Wales from 1994 to 1996. The study found concordance rates for ASD was 87% in monozygotic twins and 22% in dizygotic twin and concluded that liability to autism derived from substantial genetic and moderate nonshared environmental influences (Colvert et al., 2015). Nordenbaek et al. (2014), used data from the nationwide Danish Twin Registry, to conduct a genetic epidemiological study of autism based on 7,296 same sexed twin pairs aged 3 to 14 years. A clinical assessment, diagnostic interview, observation and cognitive examination was performed on 13 monozygotic twin and 23

dizygotic twin pairs. The concordance rate for ASD was found to be 95.2% in monozygotic twins and 4.3% in dizygotic twin. The high monozygotic and low dizygotic concordance rate suggested the heritability of autism (Nordenbaek, Jorgensen, Kyvik, & Bilenberg, 2014). Studies of twins can differentiate genetic versus environmental effects and have thus been a powerful tool in providing evidence of the importance role of genetic factors in ASD (Nordenbaek et al., 2014).

Although autism is highly heritable (Tick, Bolton, Happe, Rutter, & Rijdsdijk, 2016), no single pattern of inheritance (either autosomal dominant or recessive) is observable within families (Howes, Rogdaki, Findon, Wichers, & Charman, 2018). In the majority of cases of autism, the genetic risk is polygenic, involving multiple single nucleotide polymorphisms (SNPs), each of minor effect (Clarke et al., 2015; Gaugler et al., 2014). Single nucleotide polymorphisms are the most common type of genetic variation among individuals (U.S. National Library of Medicine [NLM], 2018). They can act as biological markers and also be used to track the inheritance of disease carrying genes within families (NLM, 2018). Based on existing literature, Gaugler et al. (2014) showed that SNP-based heritability, called common inherited variations, accounted for 49% of autism liability; rare inherited variations accounted for 3%; and de novo variations, which arise spontaneously before or shortly after fertilisation (Daly, 2017), accounted for 3% of autism liability. A combination of both common and unique environmental factors accounted for the remaining 41% of autism liability (Gaugler et al., 2014).

Environmental factors. A number of environmental factors – preconception, prenatal, perinatal, and postnatal – have been hypothesised to influence the emergence and development of ASD (Mandy & Lai, 2016). Environmental factors can be considered any factors that are not genetic or inherited (Mandy & Lai, 2016). Many of these risk factors are outlined below.

Preconception environmental risks. Preconception environmental risk factors that have been identified include advanced parental age at the time of a child's birth (Gardener, Spiegelman, & Buka, 2009; Wu et al., 2017), and a short interpregnancy (i.e., interpregnancy interval of less than 12 months) or long interpregnancy interval (i.e. interpregnancy interval of more than 60 months)

(Cheslack-Postava et al., 2014). Each of these risk factors have been identified in a number of studies of children with ASD (Hultman, Sandin, Levine, Lichtenstein, & Reichenberg, 2011; Sandin et al., 2012; Cheslack-Postava et al., 2014; Durkin, DuBois, & Maenner, 2015).

Prenatal and perinatal factors. Prenatal risk factors associated with increased risk of ASD include air pollution, *in utero* valproate exposure, maternal gestational bleeding, maternal infections and fever during pregnancy, maternal diabetes and maternal prenatal psychiatric medication or psychoactive drugs use (Atladdottir et al., 2010; Becerra, Wilhelm, Olsen, Cockburn, & Ritz, 2013; Christensen et al., 2013; Gardener et al., 2009; Jiang et al., 2016; Jung, Lin, & Hwang, 2013; Kalkbrenner et al., 2018; Lam et al., 2016; Lee et al., 2015; Talbott, Marshall, Rager, Arena, Sharma, & Stacy, 2015; Volk, Hertz-Picciotto, Delwiche, Lurmann, & McConnell, 2011; Xiang et al., 2015; Zerbo et al., 2013; 2015). Perinatal and neonatal factors found to be associated with ASD include birth complications such as abnormal foetal presentation, umbilical-cord complications, foetal distress, birth injury or trauma, low birth weight, congenital malformation, respiratory distress or no breathing after birth, and medical intervention in the first month (Gardener, Spiegelman, & Buka, 2011). The authors suggested that the observed association between perinatal and neonatal factors and risk of autism might be influenced by and/or operate in combination with pre-existing prenatal factors (Gardener et al., 2011).

Gene-Environment interplay - interaction and correlation. Genetic and environmental factors are not simply additive (Mandy & Lai, 2016) and are best conceptualised as two broad forms of gene-environment interplay – interaction and correlation (Rutter, 2006; Rutter et al., 1997). *Gene-environment interaction* refers to the fact that genetic factors can be modified in the presence of specific environmental factors or the effects of the environment are influenced by the existence of a specific genetic predisposition (Rutter, 2006). *Gene-environment correlation* refers to the indirect effects of genetic operating through specific environment (Mandy & Lai, 2016). Hence, phenotypic variances of behavioural or clinical characteristics of ASD can be analysed by looking at the contribution of genetic and environmental factors, and the gene-environment interplay

(Chaste & Leboyer, 2012; Kim & Leventhal, 2015; Mandy & Lai, 2016; Meek, Lemery-Chalfant, Jahromi, & Valiente, 2013). Based on the findings from an empirical study that siblings unaffected by ASD had fewer prenatal and perinatal complications than their affected siblings, some level of interactions existed between genetic background and environmental factors such that siblings with ASD might react differently to the same environmental stimuli and might have less tolerance to the prenatal experience compared with their siblings (Glasson et al., 2004, as cited in Chaste & Leboyer, 2012). Using a developmental psychopathology framework to synthesise literature focusing on gene-environment interaction and correlation, Mandy and Lai (2016) suggested gene-environment interplay as a possible interpretation for findings such as advanced paternal and maternal ages might independently increase offspring risk for ASD, in utero valproate exposure might serve as a causal risk factor for ASD, prenatal folate intake seemed to operate as a gene-environment interplay by providing the strongest protective effect for mothers and children who have a genotype associated with inefficient folate metabolism.

Challenging Behaviour in Children With ASD

Challenging behaviours are commonly defined as those behaviours that are physically dangerous, not socially acceptable, and that have a negative impact on education or living placement (Matson, Mahan, Hess, Fodstad, & Neal, 2010). It is estimated that approximately 6.0% to 9.3% of typically developing preschool aged children exhibit some form of challenging behaviour (Briggs-Gowan, Carter, Skuban, & Horwitz, 2001). By comparison, 95% percent of children aged 2 to 18 with ASD exhibit at least one form of challenging behaviour (Matson, Wilkins, & Macken, 2009; Jang, Dixon, Tarbox, & Granpeesheh, 2011). In children with autism, challenging behaviours that are commonly observed include unusual eating habits, sleep problems, tantrums, aggression, stereotypy, property destruction, and self-harm (Dominick, Davis, Lainhart, Tager-Flusberg, & Folstein, 2007; Jang et al., 2011; Matson et al., 2009). Challenging behaviors in children with ASD are highly persistent if untreated, and are commonly reported not only in

childhood, but also adolescence and adulthood (Bauminger, Solomon, & Rogers, 2010; Conroy, Dunlap, Clarke, & Alter, 2005).

Externalising behaviours. Externalising behaviours are negative behaviours toward others and include tantrums, defiance, noncompliance, and aggressive behaviours (Sellinger & Elder, 2016; Farmer et al., 2015). Two studies that each included over 1000 children with ASD between the ages of 2 and 17 found that approximately 54% respondents were currently engaging in some form of aggressive behaviour (Kanne & Mazurek, 2011; Mazurek, Kanne, & Wodka, 2013). In a further study, including 414 children with ASD aged 1 to 21 years old, Farmer et al. (2016) found that 50% of those with ASD engaged in at least one form of challenging behaviour as measure on the Children's Scale for Hostility and Aggression: Reactive/Proactive (C-SHARP). This rate is far in excess of what is observed among children with typical development (Briggs-Gowan, Carter, Skuban, & Horwitz, 2001).

Several researchers have suggested that there is an association between externalising behaviours problems and core ASD symptoms whereby, core autism symptoms such as difficulties related to social function (i.e. interpreting social cues and understanding expectations) contribute to rates of externalising behaviours (Shea, Payne, & Russo, 2018; Volker et al., 2010). Konst et al. (2013) suggest that as the severity of core symptoms increases, communication, social skills, and comprehension abilities might be affected leading to an increased likelihood of externalising behaviour problems. Other co-occurring problems commonly observed in children with ASD, such as sleep and sensory problems, are also thought to exacerbate the likelihood of externalising behaviour problems (Mazurek et al., 2013).

Internalising behaviours. Internalising behaviour refers to behaviour that is internal and not observable and reflects emotional or psychological state (Liu, Chen, & Lewis, 2011). Depression and anxiety disorders are two of the most frequent comorbid conditions in autism population (Ghaziuddin, Ghaziuddin, & Greden, 2002; White, Oswald, Ollendick, & Scahill, 2009). White et al. (2009) conducted a literature review exploring the association between autism and

anxiety and found that between 11% and 84% of children with autism showed signs of clinically significant anxiety. In comparison, anxiety disorders in typically developing children were estimated to be between 2.2% and 27% (Costello, Egger, & Angold, 2005). Some researchers suggested that anxiety might be inherent in autism due to the presence of social avoidance, deficits in social awareness and experience, or heightened sensitivity to environmental stimuli and sensory modulation dysfunction (Chalfant, Rapee, & Carroll, 2007; Storch et al., 2013; White et al., 2009).

Recent research suggests that 13% to 44% of children with ASD meet diagnostic criteria for depression (Leyfer et al., 2006; Salazar et al., 2015; Strang et al., 2012). There are a number of plausible explanations for why children with ASD experience increased rates of depression, including the fact that social deficits inherent in children with ASD might aggravate depression symptoms in socially challenging environments (Gadow, Guttman-Steinmetz, Rieffe, & DeVincent, 2012). In addition, due to the social and communication deficits, children with autism might avoid group interactions, experience difficulty in expressing themselves and seeking support, which may lead to symptoms of depression (Andersen, Skogli, Hovik, Egeland, & Oie, 2015).

Sleep and Children With ASD

The function of sleep. Sleep is a critical component of healthy development and is required for children's physical and mental health (Chaput et al., 2016; Hong, Kahn, & Perez, 2017). The negative impact of poor sleep on behaviour and cognition in typically developing children is well recognised (Chawla, Burgess, & Heussler, 2018). There is, however, a lack of consensus concerning the function of sleep (Krueger, Frank, Wisor, & Roy, 2016). There is substantial evidence to suggest that sleep has an important role in preserving calories, immune enhancement, restoring brain energy, neuronal and neuroglial connectivity, and performance restoration (Krueger et al., 2016).

One way to determine the function of sleep is to conduct sleep-restriction experiments (Wiggs, 2007). A number of studies have conducted sleep restriction experiments with children and have found that this has a negative effect such as an increase in parent-reported oppositional and

inattentive behaviour, a decrease in positive mood, behavioural/emotional problems at older age, inability to take full advantages of positive experiences, less adaptive in challenging contexts, and an inability to manage emotion regulation effectively (Berger, Miller, Seifer, Cares, & Lebourgeois, 2012; Fallone, Owens, & Deane, 2002; Gregory & O'Connor, 2002). Given the potential impact of sleep deprivation on optimal cognitive and emotional functioning, it is important that sleep problems are addressed properly.

Sleep problems and children with ASD. One of the most commonly reported challenging behaviours experienced by children with ASD is sleep problems (Dominick et al., 2007; Levin & Scher, 2016; Wiggs & Stores, 2004; Williams, Sears, & Allard, 2004). Prevalence rates of sleep problems in children with ASD range from 44% to 89%. (Cotton & Richdale, 2006; Hodge et al., 2014; Krakowiak et al., 2008; Liu, Hubbard, Fabes, & Adam, 2006; Wiggs & Stores, 2004). This compares to 11% to 50% of typically developing children (Cotton & Richdale, 2006; 2010; Hodge et al., 2014; Krakowiak et al., 2008). Several possible reasons for such a wide variation in terms of prevalence rates are variations in the definition of sleep problems, different instruments used to measure sleep problems, and age-related changes in sleep problems (Hodge et al., 2014; Krakowiak et al., 2008). Compared to typically developing children, sleep problems in children with ASD were more likely to persist or increased marginally as the children with ASD grew older (Hodge et al., 2014).

Trajectory of sleep problems in children with ASD. Unless treated effectively, sleep problems often last for many years, sometimes continuing into adulthood (Baker & Richdale, 2015; Cohen, Conduit, Lockley, Rajaratnam, & Cornish, 2014; Cortesi et al., 2010; Davis et al., 2012; Ming, Brimacombe, Chaaban, Zimmerman-Bier, & Wagner, 2008; Robinson & Richdale, 2004; Tani et al., 2004). Wiggs and Stores (2004) reported that 63% of children with an autism diagnosis and sleep problems experienced persistence of sleep problems over time. In a study to identify age-related changes in multiple domains of the sleep of children with autism versus changes in typically developing children, Hodge et al. (2014) reported significantly poorer sleep quality and quantity in

children with autism, particularly around the age of 6 to 9 years old. The rates of sleep difficulties were higher for the older children with autism than the younger children, 87.5% versus 84%, respectively. In contrast, within typically developing children, the rates of sleep difficulties were lower for the older children than the younger children, 37.5% versus 72%. Similarly, in a longitudinal study by Verhoeff et al., (2018), it was found that sleep problems tended to decrease over time for typically developing individuals yet increase among those with traits of ASD. Given that sleep problems in children with autism are consistently associated with impaired daytime functioning and well-being, it is crucial that effective interventions are provided at a young age (Cuomo et al., 2017).

Types of sleep problems in children with ASD. It is impossible to describe all sleep disorders occurring in children with autism within the scope of this thesis. Hence, the discussion is confined to those sleep disorders most commonly reported among this group of children. The most inclusive categories that had been used to define childhood sleep disorders were dyssomnias and parasomnias (Schreck & Mulick, 2000)

Dyssomnias. A greater frequency of dyssomnias was found in children with ASD, as compared with children with intellectual disability, children with specific developmental problems and typically developing children (Schreck, 1997, as cited in Richdale, 1999). In general, dyssomnias refers to difficulty falling asleep, frequent night wakings, or excessive sleepiness (Schreck & Mulick. 2000; Wirojatan et al., 2009).

Sleep onset insomnia. The most common types of sleep problems in children with autism problems associated with sleep onset and maintenance (Cortesi et al., 2010; Malow et al., 2014; Souders et al., 2009). Children with autism have difficulties in sleep initiation, shorter sleep time, and more interrupted sleep patterns when compared to their typically developing peers (Elrod & Hood, 2015; Miano et al., 2007). Sleep onset insomnia can include the sleep-onset association type or limit-setting type. Children with the sleep-onset association type of behavioural insomnia typically depend on a specific stimulus, object, setting, or person to initiate sleep or to resume

normal sleep (Souders et al., 2017). Children with limit-setting type of behavioural insomnia struggle or refuse to sleep at bedtime because of difficulties dealing with limits set by the caregiver. These resistance or refusal behaviours often escalate each night and eventually become disruptive (Souders et al., 2017).

Sleep maintenance problems. Sleep maintenance problems in children with autism include frequent night wakings and early morning wakings (Giannotti et al., 2008). Parents report prolonged night wakings lasting for up to 2-3 hours wherein the child might get up, laugh, talk, play with toys or various objects, and run around (Cortesi et al., 2010; Patzold, Richdale, & Tonge, 1998). Due to the difficulty in initiating and maintaining sleep, children with autism might have difficulty establishing appropriate sleep-wake cycles and might experience circadian rhythm sleep disorders, such as delays in falling asleep, waking early in the morning, feeling tired when awake, and feeling sleepy during the day (Taylor, Schreck, & Mulick, 2012).

Parasomnias. Parasomnias refers to abnormal behaviours associated with sleep such as sleepwalking and disorders related to incomplete dissociation of wakefulness from sleep (Howell, 2012). Parasomnias, such as nightmares and sleepwalking, are less frequently reported in children with autism (Williams, Sears, & Allard, 2004). Findings on the prevalence of parasomnias in autism are in fact, relatively mixed. Two studies have reported low incidence rates of parasomnias among children with ASD (Patzold, Richdale, & Tong, 1998; Williams et al., 2004). In contrast, Schreck and Mulick (2000) reported that children with ASD exhibited higher rates of parasomnias when compared to typically developing children or children with developmental disabilities or pervasive developmental disorder. Liu et al. (2006) also found that all sleep problems, including parasomnias, were very prevalent in children with ASD and suggested the possibility of compounded effects of multiple sleep problems.

Sleep-disordered breathing. Sleep-disordered breathing encompasses a spectrum of sleep-related breathing disorders, including snoring, obstructive sleep apnoea, central sleep apnoea, sleep-

related hypoventilation and respiratory-related arousals (Mohammadieh, Sutherland, & Cistulli, 2017).

Obstructive Sleep Apnoea. Obstructive sleep apnoea refers to conditions in which frequent collapse of upper airway during sleep causing recurrent disturbances in gas exchange which lead to oxygen desaturations, nocturnal arousal and fragmented sleep (Jordan, McSharry, & Malhotra, 2014; Mohammadieh et al., 2017). One of the most common symptoms of Obstructive Sleep Apnoea in children is snoring (Owen, Canter, & Robinson, 1996). However, both snoring and obstructive apnoea are two extremes of a wide spectrum of upper airway resistance. In children with obstructive sleep apnoea, in addition to snoring, the parent or caregiver might also comment on breathing difficulties during sleep and apnoea. Nonetheless, the clinical presentation of obstructive sleep apnoea was usually very nonspecific and a more definitive diagnostic required the use of an overnight polysomnographic evaluation (Gozal, 2001). The present study focuses on behavioural sleep problems and, therefore, excludes all children with obstructive sleep apnoea.

Co-sleeping. Co-sleeping is relatively common in children with autism. The high frequency of co-sleeping might be due to sleep problems common in children with autism, parental or caregiver concerns and the intention to monitor and care for the child if awake during the night, difficulties with self-soothing and self-regulating, and difficulties in adjusting to different sleep environment (Bastida-Pozuelo, Meltzer, & Sanchez-Ortuno, 2018; Liu et al., 2006). By being present at bedtime to reduce sleep onset latency in children with autism, a parent might become a negative sleep onset association requiring full night co-sleeping (Bastida-Pozuelo et al., 2018). Evidence of an association between co-sleeping and behavioural sleep problems are not clear (Mileva-Seitz et al., 2017). A recent longitudinal population-based study of 4,231 children from birth until the age of six years found an association between co-sleeping and impaired child mental health at the age of six years (Santos et al., 2017). In contrast, an 18-year longitudinal study of 205 families found no association between co-sleeping and behaviour problems and difficulties in peer

and intimate relationships at age of 6 years and age of 18 years old (Okami, Weisner, & Olmstead, 2002).

Etiology of sleep problems in children with ASD. Spielman et al. (1987) proposed an insomnia model which describes predisposing, precipitating, and perpetuating factors crucial to the development and maintenance of insomnia. This includes genetic, physiological, behavioural, psychological, and environmental factors (Spielman, Caruso, & Glovinsky, 1987). Predisposing factors, such as the neurobiological and neuropsychiatric vulnerability of children with autism (Souders et al., 2017), provide differential susceptibility to individuals. Precipitating factors push an individual over a hypothetical threshold to produce acute symptoms of insomnia. These precipitating factors could include environmental stressors such as changes in sensory stimuli and sleep routine, psychological stressors such as difficulties encountered during the daytime, or physiological stressors such as sickness (Souders et al., 2017). Perpetuating factors prevent the individual from resuming normal sleep. Perpetuating factors, such as parental presence until sleep onset or during a daytime nap, could subsequently be introduced by caregivers. Such behavioural conditions that are initiated by caregivers to relieve symptoms of insomnia in their children often inadvertently worsen the symptoms and perpetuate insomnia long-term (Souders et al., 2017).

Biological or co-morbidities or medical factors. Biological or medical factors that are associated with sleep disorders in ASD include abnormalities in neurotransmitter systems (e.g., melatonin, Gamma-Aminobutyric Acid (GABA), and serotonin); medical conditions that disrupt sleep (e.g., gastrointestinal (GI) dysfunctions, Obstructive Sleep Apnoea, and epilepsy); and the medications taken to treat these conditions (Goldman et al., 2009; Kothare & Kotagal, 2011; Liu et al., 2006; McCue, Flick, Twyman, & Xian, 2017). Abnormalities in melatonin secretion are also widely reported in earlier studies of sleep in children with autism (Melke et al., 2008; Tordjman et al., 2015). Melatonin is a neurohormone that regulates the circadian sleep-wake rhythms (Arendt, 2003; Tordjman et al., 2013).

Medical conditions that disrupt sleep have also been reported in several studies. Significant associations have been found between sleep problems and GI dysfunction among children with autism (Kang, Wagner, & Ming, 2014; Maenner et al., 2012; Mannion, Leader, & Healy, 2013; Ming et al., 2008). In one of the largest studies to estimate the relationship between GI dysfunction and sleep problem severity in children with ASD, McCue et al. (2017) found that children with autism who had GI dysfunctions had a significantly higher probability of having sleep disorders and were twice as likely to experience multiple sleep problems. Several studies have also reported a reduction in sleep problems and features of autism following the treatment of medical conditions that disrupt sleep. For example, Malow et al. (2006b) reported improvements in autism features, such as social communication, repetitive and restricted behaviours, attention, hypersensitivity, and sleep after treatment for Obstructive Sleep Apnoea in a 5-year-old female with autism. In a more recent study, Murata et al. (2017) reported significant improvement in the features of autism after Obstructive Sleep Apnoea treatment in 30 children with autism. Other medical conditions that disrupt sleep in children with autism include sleep movement disorders (e.g., restless leg, bruxism, stereotypic movements), daytime sleepiness, and epilepsy (Liu et al., 2006; Rodriguez, 2007; Schreck & Mulick, 2000).

Medications, such as psychostimulants and atomoxetine, taken to treat psychopathology or the medical conditions, described above, might also disrupt sleep in children with autism (Liu et al., 2006; Malow et al., 2012; Mayes & Calhoun, 2009; Singh & Zimmerman, 2015). Some medications result in side effects such as sedation or insomnia, or long term neurological symptoms and disorders (i.e. tardive dyskinesia) (Matson & Hess, 2011; McCue et al., 2017).

Psychological causes. Externalising behaviour, such as hyperactivity, aggression, oppositional behaviour, impulsivity, explosiveness, attention, and impulsivity have been related to sleep problems in children with autism (Allik, Larsson, & Smedje, 2006; Hollway & Aman, 2011; Malow et al., 2006a; Mayes & Calhoun, 2009). In addition to externalising problems, studies reported noticeable affective and emotional problems (i.e., anxiety and depressive symptoms)

between autism good and poor sleepers (Allik et al., 2006; Hollway & Aman, 2011; Malow et al., 2006a) suggesting that emotional difficulties may play a role in the development of insomnia in children with ASD (Hollway & Aman, 2011; Malow et al., 2006a).

Social or familial factors. Social or familial factors may also contribute toward sleep problems in those with ASD. This includes problematic sleep-related cognitions, active parental involvement and not allowing children to self-soothe. For example, problematic sleep-related cognitions (i.e., doubts about parenting competence and difficulties with limit setting) expressed by parents or caregivers were associated with disrupted sleep in both typically developing or children with autism. Likewise, active parental or caregiver involvement, as opposed to allowing children to self-soothe, during transitions from awake to asleep was associated with shorter and more fragmented sleep in children with autism (Levin & Scher, 2016). Nevertheless, empirical support that linked symptoms of dysregulation, such as sleep problems and somatic complaints, to parent behaviours was limited and no study investigated how parent behaviours influence the development of dysregulation among children with autism (Wiggins et al., 2018). A more recent study investigating the presence of sub-clinical autism characteristics (i.e., broader autism phenotype) in families of children with autism found that the presence of broader autism phenotype in parents of children with autism was significantly associated with the child having anxiety, depression and sleep problems (Rubenstein et al., 2018).

Environmental factors. Behaviours that promote quality sleep and longer sleep duration (i.e. sleep hygiene) play an important role in sleep development and sleep problems in early childhood (Sadeh, Tikotzky, & Scher, 2010). Sleep hygiene factors that might contribute toward sleep problems in children with autism include issues with the bedroom environment (e.g., television, environmental light, loud noises, and temperature), sleep scheduling (e.g., inconsistent bedtimes, wake up time and nap times, bedtime routine), food and beverage consumption (e.g., food intake immediately prior to bed, caffeinated drinks) and access to highly arousing pre-bedtime activities (e.g., use of electronic media devices, such as tablets and mobile phones, exercise

immediately prior to bed). Each of these factors can act to precipitate or perpetuate sleep disturbances in children with ASD (Akacem, Wright, & LeBourgeois, 2016; Mazurek, Engelhardt, Hilgard, & Sohl, 2016; Mindell, Meltzer, Carskadon, & Chervin, 2009; Richdale & Schreck, 2019; van der Heijden, Stoffelsen, Popma, & Swaab, 2018).

A behavioural model of sleep disturbance. Sleep is neither respondent nor operant behaviour. It is not a respondent behaviour because it is not possible to consistently elicit sleep by presenting the unconditioned stimulus for sleep. It is also not operant behaviour because it is not possible to increase and decrease the frequency and duration of sleep by reinforcing or punishing sleep (Blampied & France, 1993). Instead, it is a biobehavioural state in which both biological and behavioural variables are involved in the initiation and maintenance of sleep. A behavioural model of sleep must, therefore, incorporate the relationship between sleep and the variables that are critical for both respondent behaviour (conditioned and unconditioned stimuli) and operant behaviour (three-term contingencies involving antecedent stimuli, behaviours, and response consequences) (Blampied & France, 1993). Although sleep in itself is a state and not a behaviour per se, falling asleep is an operant behaviour performed to produce sleep and reinforced by sleep itself (Bootzin, 1977, as cited in Blampied & France, 1993). In extending this argument further, Ferster et al. (1975) suggested an operant behaviour chain that began with bed-preparation behaviours and ended with a period of behavioural quietude just before falling asleep. To enable sleep to occur reliably and appropriately, Bootzin (1977) assumed that the operant behaviour chains needed to come under the control of appropriate discriminative stimuli. Each component of the behaviour chain might have different discriminative stimuli and each component might provide the discriminative stimuli for the subsequent component of the chain. A period of behavioural quietude enables sleep-preparatory cues to be discriminated and facilitate falling asleep. In contrast, continued activity will compete and prevent any sleep-preparatory cues to be discriminated and interfere with falling asleep. The second key assumption made by Bootzin (1977) was that sleep difficulties might be associated with inappropriate and sleep-competing behaviour. These competing behaviours were maintained by

various positive and negative reinforcers and might also come under the control of discriminative stimuli. Based on the behavioural analysis of sleep mentioned above, Blampied and France (1993) proposed two important aspects encompassing the antecedents and consequences associated with the bedtime behavioural chain. First, appropriate discriminative stimulus control needed to be available for each component of the behavioural chain. Second, the bedtime behavioural chain and sleep-promoting behaviours needed to be shaped, strengthened and maintained by contingencies of reinforcement. Absence of appropriate discriminative stimulus control might contribute to bed refusal and delay in sleep initiation. Furthermore, distinctiveness and consistency of these discriminative stimuli might affect the regularity and orderliness with which the bedtime behaviour chain is started and completed. These discriminative stimuli could include bed-related cues and self-produced comfort cues. However, some external cues (e.g., bedtime feeding, pre-bed, and pre-sleep rituals) might facilitate falling asleep initially, but might interfere with falling asleep once habitual. In addition, behaviour incompatible with going to bed and falling asleep also competed with sleep-compatible behaviour. The response competition from sleep-compatible and incompatible behaviour can be explained by analysis of the contingencies of positive and negative reinforcement (Blampied & France, 1993). For children, parental attention was a powerful reinforcer that was more immediate and, therefore, more potent than the reinforcer of sleep. For parents, not having a child crying and going to sleep at night was likely to be a powerful negative reinforcer. These consequences were mutually reinforcing and maintained sleep-incompatible behaviour and contributed to the other half of bed refusal and delay in sleep initiation issues mentioned above (Blampied & France, 1993). For example, children must learn behavioural quietude, allow the naturally occurring bed cues to facilitate falling asleep and subsequently reinforced by sleep. In case of sleep-onset delay, children might initially struggle with the period of behavioural quietude and will display distress behaviour in an attempt to avoid or escape from such a situation. Any distress behaviour is doubly reinforced if it is effective in gaining parental attention. Such behaviour is negatively reinforced by escape from the behavioural quietude prior to falling asleep and positively

reinforced by immediate parental attention. Both the distress behaviour and subsequent parental interaction compete with falling asleep under natural stimulus control and thus aggravating the problem. Moreover, such distress behaviour might occur earlier and earlier, while parental attention will be gained more and more immediate, resulting in prolonged bedtime tantrums and sleep initiations (Blampied & France, 1993). Similarly, in cosleeping, a practice where the child is allowed to sleep in the parents' bed during sleep initiation or upon night wakings, appropriate stimulus control could not be developed and contingencies of positive and negative reinforcement were involved in maintaining the practice (Blampied & France, 1993). Night wakings might also be maintained by similar contingencies. While growing up, an infant is likely to cry in response to waking and the parent will attend almost immediately since the immediate parental attention may shorten the infant's crying. Hence, it might be very difficult for parents to withhold their attention in order for their child to learn to resume sleep without parental involvement or interaction.

An assessment of both the clear discriminative stimulus control and contingencies of reinforcement that may shape and maintain either appropriate or disturbed sleep is crucial so that appropriate natural cues become the controlling stimulus for falling asleep and sleep-compatible behaviours are reinforced (Blampied, 2013; Blampied & France, 1993).

Behavioural factors. Behavioural causes of insomnia (e.g., poor sleep habits) might stem from the core behavioural deficits associated with autism which might impede the establishment of consistent bedtime routines and habits (Malow et al., 2014). For example, due to deficits in communication skills, children with autism might not readily understand bedtime expectations. Furthermore, children with autism might have difficulty transitioning from daytime activities and calming themselves in preparation to sleep (Malow et al., 2014).

Biopsychosocial causes. According to Richdale and Schreck (2009) sleep problems in children with autism can be the result of a combination of biopsychosocial factors. Specifically, they emphasise the complex interaction between biological, psychological or behavioural, and environmental factors in sleep problems in children with autism. For example, in children with

autism any combination of circadian rhythm dysfunction, reinforcement for sleep interfering behaviour, fears or worries about bedtime, or sensitivity to environmental stimuli might interact to exacerbate or maintain sleep problems (Souders et al., 2009; Wiggs & Stores, 2004). In addition, children with autism are more vulnerable to sleep problems due to prevalent symptoms of hyperactivity, Attention Deficit Hyperactivity Disorder (ADHD), oppositional defiant disorder (ODD) (Allik et al., 2006; Liu et al., 2006; Malow et al., 2006a) and comorbid anxiety and depression (Leyfer et al., 2006).

Bidirectional interaction between biopsychosocial cause and sleep problems. Hollway and Aman (2011) have proposed a bidirectional theoretical framework for predicting sleep disturbance in children with autism in which vulnerability factors might be associated with sleep problems and sleep problems might negatively affect or worsen the biopsychosocial risk or vulnerability factors. Intellectual functioning has been found to be a moderator of autism symptom severity and ultimately of insomnia in those with autism. Features of autism acted as vulnerability factors and predisposed children to insomnia in the presence of environmental stressors. In addition, according to the model, each of the autism core symptoms – communication deficits, restricted and repetitive behaviour, and social skills deficits – served additively to increase maladaptive coping mechanisms such as internalising, externalising, and over-arousal by affecting sleep. Hollway and Aman (2011) also found that comorbid medical conditions (epilepsy, GI problems) were associated with sleep disturbance. Finally, a bidirectional component was included to show the effect that lack of sleep had on maladaptive responding (Hollway, Aman, & Butter, 2013). In the general population, a bidirectional relationship between emotional problems and sleep problems had been hypothesised (Mulraney, Giallo, Lycett, Mensah, & Sciberras, 2016). Several studies had found that emotional problems predicted the onset of sleep problems and that sleep problems predicted the onset of emotional problems (Baglioni et al., 2011; Jansson-Frojmark & Lindblom, 2008; Johnson, Roth & Breslau, 2006).

It should also be noted that the Hollway et al. (2013) study was cross-sectional not longitudinal and, hence, directionality and causality cannot be determined (Mazurek & Sohl, 2016). In order to determine causal factors, it is important that predictions are made using both baseline and follow-up data (Shui, Katz, Malow, & Mazurek, 2018). In a longitudinal study, aimed at identifying the bi-directionality of the relationship between sleep problems and ASD, Verhoeff et al. (2018) took a population-based sample of 5,151 children who had sleep problems and autistic traits. From 1 ½ years onward they assessed sleep problems and autistic traits when the children were 1 ½ years, 3 and 6 years old. In addition, sleep problems were assessed again at the age of 9 years old. Children's sleep problems were assessed using the Sleep Problem Scale, a pre-defined Problem Scale of the CBCL (Achenbach & Ruffle, 2000). Results showed a significant long-term association between autistic traits at age 1.5 and 3 years and more sleep problems at 6 years old. That is, children either with autistic traits or with ASD at 6 years old, had more sleep problems at 9 years old. Importantly also, no evidence was found to indicate that sleep problems preceded the development of autistic traits in toddlerhood. The authors concluded that sleep problems did not precede and worsen autistic behaviour but rather co-occur with autistic traits in early childhood and increase over time in children with autism (Verhoeff et al., 2018)

Sleep problems and autism symptoms and severity. The association between the severity of autism symptoms and reduced night sleeping has been reported in some research studies (Adams Matson, Cervantes, & Godlin, 2014; Allik et al, 2006; Cohen et al., 2017; Hoffman et al., 2008; Goldman et al., 2011; Mayes & Calhoun, 2009; Patzold et al., 1998; Schreck et al., 2004; Tudor, Hoffman, & Sweeney, 2012). Schreck et al. (2004) used the Gilliam Autism Rating Scale (GARS; Gilliam, 1995) to measure core autism features and found that shorter sleep duration predicted GARS total scores, including Stereotyped Behaviour, Social Interaction, and Total Autism Quotient and the GARS Communication domain scores predicted night wakings and sensitivity to stimuli in the environment. Furthermore, in a longitudinal study using a dataset of over 50,000 nights of sleep obtained from a sample of children with low functioning autism, Cohen et al. (2017) reported that

sleep deficits increase with autism symptom severity. Mayes and Calhoun (2009) used the Pediatric Behaviour Scale (PBS) to assess both sleep problems and behaviour in a sample of 477 children with autism and reported that sleep disturbances were significantly correlated with both internalising and externalising behaviours, as measured on the PBS.

Sleep problems and social interaction and communication difficulties. Research has consistently demonstrated that sleep problems are associated with, or predictive of, difficulty developing peer relationships, social interaction problems, and communication difficulties (Schreck et al., 2004; Tudor et al., 2012; Veatch et al., 2017). Schreck et al. (2004) reported that, among fifty-five 5- to 12-year-old children with autism, shorter duration of sleep partially predicted social interaction problems and severity of stereotypic behaviours whereas over-sensitivity to stimuli in the environment was associated with communication problems and developmental abnormalities commonly related to autism (e.g., repetitive behaviours, regression in skill, delays in speech development). Consistent with Schreck et al. (2004), in a study of 335 children with autism and IQs in the normal range, Taylor et al. (2012) found children who slept more on average per night without waking and were less bothered by environmental stimuli in their sleeping environment showed better ability to effectively communicate with others during the day.

Sleep problems and stereotyped and repetitive behaviour. The results from research studies that investigated the relationship between the intensity of restricted and repetitive behaviours and sleep problems in children with autism were mixed and generally reported that sleep problems were related to the presence of higher levels of restricted and repetitive behaviours (Goldman et al., 2009; Schreck et al., 2004), although this relationship might be moderated by the level of cognitive ability (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005). In a study of 109 children with ASD aged 3 to 18 years old, Tudor et al. (2012) found that sleep onset delay was a predictor of stereotyped behaviour. In another study with 2,714 children, Veatch et al. (2017) reported that shorter sleep duration was strongly associated with an adherence to restricted and repetitive behaviours and maladaptive behaviours. In contrast, Malow et al. (2006) did not find any association between restricted and

repetitive behaviour and sleep problems, although the authors suggested that additional studies in ASD relating to sleep and daytime behaviours would be needed to validate the findings and interventional studies will be necessary to determine causality in relation to sleep and behaviour.

Sleep problems and adaptive behaviour. Adaptive behaviour is generally defined as an individual's ability to translate cognitive ability into real-life skills such as conversing with and understanding others, participating in group and community activities, interacting with others and developing relationships, taking care of one's health, grooming and domestic chores (Klin et al., 2007; Sparrow & Cicchetti, 1985). There has been limited research into the effects of sleep problems on adaptive behaviour in children with autism (Taylor et al., 2012). In one of the first studies to investigate the impact of sleep problems on the ability to complete daily living activities, Taylor et al. (2012) reported that those children who slept less on average both at night and during the day had more difficulty with all adaptive behaviour tasks, especially motor and social skills. A more recent study by Cohen et al. (2017) that used hierarchical cluster analysis, based on an unprecedentedly large dataset of over 50,000 nights sleep for 106 children and adolescence aged between 5 and 18 years old, who had low functioning autism, also reported that children with reduced sleep duration, early morning wakings and irregular sleep timing exhibited greater impairments in adaptive functioning, as measured by the Vineland Adaptive Behaviour Scales (VABS; Sparrow, Balla, & Cicchetti, 1984). Furthermore, Krakowiak et al. (2008) reported that, lower adaptive function was associated with increased difficulties with sleep onset, night waking, and shorter duration of sleep. However, adaptive functioning difficulties did not predict the severity of either type of sleep problem or the duration of sleep (Krakowiak et al., 2008).

Sleep problems and externalising behaviour. Sleep problems in children with ASD have been strongly linked with rates of aggression, self-injury, tantrums, and hostility/irritability (Adams, Matson, & Jang, 2014; Allik et al., 2006; Bruni et al., 2007; Cohen et al., 2018; Goldman et al., 2011; Mazurek & Sohl, 2016; Patzold et al., 1998; Shui et al., 2018). In a sample of 1,784 children with autism, Goldman et al. (2011) found that poor sleepers (as reported by parents) demonstrated

greater challenging behaviour, including aggression and self-injurious behaviour, than good sleepers. Mazurek and Sohl (2016) reported that, of the different types of sleep problems, night wakings were strongly and positively correlated with physical aggression and hostility/irritability. A recent longitudinal study based on a sample of 67 children aged 6.6 to 19 years old with low functioning autism, by Cohen et al. (2018) found that longer duration of prior sleep predicted subsequent daytime challenging behaviour such as aggression, self-injury, and tantrums. Furthermore, another longitudinal study of 1,045 children and adolescence with autism without sleep problems at baseline aged 2 to 17 years old, Shui et al. (2018) reported that aggressive behaviour at baseline might also predict poor sleep one year later.

Sleep problems and hyperactivity and inattention. A number of studies have reported an association between sleep problems and hyperactivity and inattention in children with ASD (Allik et al., 2006; Cortesi et al., 2010; Goldman et al., 2009; Kothare & Kotagal, 2011; Malow et al., 2006a; Mayes & Calhoun, 2009; Mazurek & Sohl, 2016). For example, more hyperactivity in children with autism who were poor sleepers than those who were good sleepers and typically developing children (Goldman et al., 2009; Malow et al., 2006a). Of the type of sleep disturbance, sleep anxiety was positively correlated with both irritability and hyperactivity (Mazurek & Sohl, 2016). Nevertheless, results from a longitudinal study showed that sleep disturbance did not predict hyperactivity one year later for children with high functioning ASD aged 7 to 12 years old (May, Cornish, Conduit, Rajaratnam, & Rinehart, 2015) suggesting that no bidirectional relationship between sleep problems and hyperactivity.

Sleep problems and internalising behaviour. Results from studies that investigated the relationship between internalising behaviours and sleep problems have demonstrated that sleep problems have been strongly associated with anxiety and depressive symptoms (Allik et al., 2006; Malow et al., 2006a; May et al., 2015; Patzold et al., 1998; Sikora et al., 2012). Comparing the results from the Affective subscale of the CBCL between children with autism who were poor sleepers and good sleepers, Malow et al. (2006) found that poor sleepers had significantly more

depressive symptoms. In addition, some children with autism experienced significant depressive symptoms which affected sleep (Malow et al., 2006a). In a longitudinal study investigating the predictive associations between sleep disturbance and behavioural or emotional problems, May et al. (2015) reported sleep disturbance predicted anxiety one year later although the sample was limited to 7- to 12-year-old children with high functioning autism.

Common treatments for sleep problems in children with ASD. The two most commonly used treatment methods for children with ASD are melatonin and behavioural interventions. Each of these treatment approaches is outlined below.

Melatonin. The literature indicates that melatonin is an effective intervention for sleep problems individuals with ASD (Garstang & Wallis, 2006; Gringras, Nir, Breddy, Frydman-Marom, & Findling, 2017; Guenole & Baleyte, 2011; Guenole et al., 2011; Hollway & Aman, 2011; Malow et al., 2012; Maras et al., 2018; Rossignol & Frye, 2011; Wright et al., 2011). For example, placebo-controlled trials of 11-20 children with ASD, aged 4 to 16 years old, showed improvements of longer sleep latency and increased total sleep duration under melatonin compared to a placebo (Garstang & Wallis, 2006; Wright et al., 2011). In a more recent randomized, double-blind, placebo-controlled trial conducted with children aged 2 to 17.5 years old in US ($n = 196$) and in Europe ($n = 71$), a paediatric-appropriate prolonged-release melatonin was shown to be more effective in children with ASD than placebo in increasing total sleep time and reducing sleep latency. Treatment-related adverse events such as somnolence and mood swings were reported in 11.7% and 1.7% of the treatment group, respectively. In comparison, somnolence and mood swings affected 3.1% and 6.2% of the placebo group, respectively. Severe adverse events such as agitation and fatigue were reported in 8.3% and 6.7% of the treatment group, respectively. In comparison, agitation and fatigue affected 4.6% and 3.1% of the placebo group, respectively (Gringras et al., 2017). In a subsequent follow-up study, 52 weeks later, on those children who completed the initial 13-week in Gringras' (2017) trial, paediatric-appropriate prolonged-release melatonin continued to show effectiveness in increasing total sleep time, reducing sleep latency and night wakings. Fatigue

and mood swings were reported in 5.3% and 3.2% of treatment group, respectively (Maras et al., 2018). Although severe adverse events such as fatigue had decreased from 6.7% to 5.3% of the treatment group, treatment-related adverse events such as mood swings had almost doubled from 1.7% to 3.2% of the treatment group highlighting the limitations of melatonin. Furthermore, parents of children with ASD preferred behaviourally-based interventions over medications (Williams et al., 2004).

Behaviourally-based interventions. Behaviourally-based interventions for sleep problems in children with ASD have been widely researched (Brown, Kuo, Phillips, Berry, & Tan, 2013) and can be organised into two major categories – antecedent-based and consequence-based procedures (Turner & Johnson, 2012).

Antecedent-based procedures. In young children with ASD, antecedent-based procedures have been used to address delayed sleep onset and night wakings (Delemere & Dounavi, 2018; DeLeon, Fisher, & Marhefka, 2004; Durand & Christodulu, 2004; Durand, 2002). Examples of antecedent-based procedures are bedtime fading interventions, sleep restriction procedure, and scheduled awakening procedure. In bedtime fading interventions, a bedtime is chosen closer to the time when the child is naturally fall asleep. This bedtime is observed until sleep onset occurs within a few minutes of the bedtime. Bedtime fading was found to reduce night wakings, increase total sleep duration and decrease sleep onset latency for young children with ASD (DeLeon et al., 2004; Delemere & Dounavi, 2018). In a sleep restriction procedure, parents or caregivers are requested to temporarily reduce children's total sleep time such that the child is in bed for only 90% of the total amount of usual reported sleep time. Once appropriate sleep behaviour is achieved, the sleep restriction is removed so that the total amount of sleep is aligned with a developmentally-appropriate norm. Sleep restriction appeared to result in a reduction of night wakings and elimination of sleep disturbances in young children with ASD (Durand & Christodulu, 2004). In a scheduled awakening procedure, parents or caregivers are requested to wake the child 15 minutes before the reported spontaneous time of night waking. Parents would then respond to the child as if

the night waking happens spontaneously. Over time, the scheduled awakening is gradually and systematically faded to occur at a longer interval of uninterrupted sleep until the intervals between night wakings are longer in duration. Scheduled awakening increased total sleep time for two out of three children with ASD (Durand, 2002). So far, scheduled awakenings only meets the criteria to be considered possibly efficacious with more research needed. To be considered probably efficacious or well-established interventions, more replications of existing studies are needed (Vriend et al., 2011).

Sleep hygiene. Sleep hygiene modification is another example of an antecedent based modification and has been shown to be an effective intervention for sleep problems in children with ASD. Common sleep hygiene recommendations include selecting a suitable and consistent bedtime and wake time, establishing a positive and consistent bedtime routine, and reducing emotional/behavioural stimulation at night. In addition, addressing sensory hypersensitivity associated with stimuli such as noises or tactile sensitivity to clothes and blankets in the sleep environment might help improve sleep quality (Cortesi et al., 2010). A more recent study by van der Heijden et al. (2018) reported that sleep problems in children with ASD were related to inadequate sleep hygiene while, in typically developing children, sleep problems were related to both chronotype and inadequate sleep hygiene. The authors suggested that sleep hygiene improvement might be effective in children with ASD and proposed that treatment strategies for sleep problems should focus initially on sleep hygiene improvements. However, the authors also found that the negative effects of inadequate sleep hygiene seemed to be diminished by the presence of comorbid anxiety or depression and cautioned that more research was needed to understand the interplay of sleep hygiene, anxiety/depression, and sleep problems in ASD (van der Heijden et al., 2018).

Consequence-based modifications. Consequence-based modifications involve manipulating reinforcement so as to shape and maintain a desirable behaviour and reduce undesirable behaviour. Examples of consequence-based interventions are standard extinction and modified extinction procedures (Turner & Johnson, 2012). In standard extinction procedures, parents are advised to

avoid any interaction with the child after they bid their child goodnight, unless absolutely necessary for safety reasons (Vriend et al., 2011), in order to eliminate parental attention that may be reinforcing the sleep problem (Mindell, 1999). While extinction procedures are effective, an extinction burst might occur in which negative behaviours might temporarily increase before an improvement is observed. As a result, education on extinction bursts and support given to parents during the procedure is crucial to help increase parental adherence to treatment and thereby the possibility of improvement. A modified version of extinction offers an alternative solution. Examples include the minimal check procedure in which regular checking is performed by parents or caregivers throughout an extinction procedure (France & Blampied, 2005; Richdale & Wiggs, 2005) or graduated extinction in which challenging behaviours are gradually reduced through gradual withdrawal of reinforcement over time (Singh & Zimmerman, 2015; Wiggs & France, 2000). In this modified approach, instead of eliminating interaction with the child throughout the night, parents are advised to ignore bedtime disruptions for a predetermined length of time. At the end of that time, if the bedtime disruptions persist, parents will settle the child back in bed but the interaction will be at a minimum (Vriend et al., 2011). Studies had found standard or graduated extinction to be effective for children with ASD in reducing bedtime resistance, night wakings, sleep onset delay, co-sleeping (Montgomery, Stores, & Wiggs, 2004; Reed et al., 2009; Weiskop, Matthews, & Richdale, 2001; Weiskop, Richdale, & Matthews, 2005). Furthermore, reduction in night wakings and bedtime resistance were maintained at 6- and 12-month follow-ups (Montgomery et al., 2004; Weiskop et al., 2005). To date, only extinction meet the criteria for a possibly efficacious intervention requiring more research (Schreck, 2001) and more replications are needed for interventions to be considered probably efficacious or well-established (Vriend et al., 2011).

Functional Behavioural Assessment (FBA) interventions. Given that the number of variables which initiate or exacerbate sleep problems are unique and different for each individual, understanding these individual-specific variables is crucial for effective treatment formulation and implementation. Functional behavioural assessment (FBA) is a process commonly used to identify

the antecedents (context or setting), consequences (e.g., parent attention or responses), and the function of a specified problem behaviour (the sleep problem). Treatment plans developed based on the outcomes of FBA are more effective because the intervention plans addressed the individual-specific variables that initiated and maintained the behaviour (McLay, France, Blampied, Danna, & Hunter, 2017). Despite strong evidence to support the use of FBA to inform interventions for challenging behaviour (Beavers, Iwata, & Lerman, 2013; Hanley, Jin, Vanselow, & Hanratty, 2014) and sleep problems (Jin, Hanley, & Beaulieu, 2013; McLay et al., 2017), there is very little research documenting the FBA process from assessment through to treatment (McLay et al., 2017). Given the multiple individual-specific variables that might impact sleep disturbance, information from the behaviour assessment and functional analysis part of the FBA needs to be integrated into a coherent, albeit tentative, account of the problem, and then linked with a comprehensive and individualised treatment plan that can be implemented by parents or caregivers (Blampied, 2013).

Two of the essential component of FBA informed interventions are elimination or modification of sleep-competing antecedent and consequence variables and determination of the function of the undesirable behaviour (McLay et al., 2017). Based on a sample of 3 young children, 2 of whom had been diagnosed with ASD, Jin et al. (2013) demonstrated the efficacy of individualised function-based sleep interventions for young children. Behavioural assessment was guided by the Sleep Assessment and Treatment Tool (SATT; Hanley, 2005). SATT is an open-ended interview designed to identify specific sleep problems and the environmental variables that triggers the sleep problems. Specific problems and environmental variables asked during the interview include: history of the child's sleep problems; target sleep goals (e.g., bedtime, wake-up time); identification of specific sleep problems (e.g., delayed sleep onset, night wakings, issues with sleep hygiene and bedtime routine, early morning wakings), description of antecedent conditions and consequences or responses; identification of the child's current bedtime and sleep dependencies (i.e., those events, interactions, items that appear to promote sleep); identification of possible sleep interfering behaviours and the corresponding reinforcers; and descriptions of treatment options

available to the parents or caregivers. By utilising SATT, researchers were able to formulate a hypothesis about the functions of sleep interfering behaviours and to evaluate environmental contingencies maintaining the child's sleep difficulties. Treatments include procedures to enhance the discriminative stimuli and establishing operations for behavioural quietude and to weaken the reinforcers for sleep interfering behaviours. For example, delayed sleep onset was shortened by increasing the value of sleep through adjusting the child's bedtime in accordance to developmental norms and their current sleep phases. Sleep interfering behaviours are reduced by eliminating the contingency between the sleep interfering behaviour and its reinforcers and providing access to the reinforcers prior to bedtime and independent of the interfering behaviour. Night wakings were addressed by eliminating inappropriate sleep dependencies (e.g., conditions that were available only at bedtime) and gradually establishing sleep dependencies on events that were available both at bedtime and throughout the night (Jin et al., 2013).

In another example, Friedman and Luiselli (2008) reported an effective intervention that eliminated excessive daytime sleep in a child with ASD. By using FBA, the authors hypothesised that the reported daytime sleep was maintained primarily by automatic reinforcement and might have functioned to escape or avoid nonpreferred interactions. In order to eliminate antecedent and consequence events that lead and reinforce daytime sleep, a number of procedures were combined, including removing sleep-associated stimuli; interruption and redirection to preferred and highly stimulating objects and activities upon detection of sleepiness; and positive reinforcement through positive interactions and delivering praise for appropriate behaviour alternative to excessive daytime sleep (Friedman & Luiselli, 2008).

A more recent study, utilising FBA to treat sleep problems in a child with ASD, by McLay et al. (2017) reported a reduction in the frequency of curtain calls (i.e. behaviours intended to avoid or delay going to bed), reduction in frequency and duration of night wakings, reduction in bedtime resistance, and reduction in sleep onset delay. Based on FBA, multiple antecedent and consequence variables were identified as contributing towards sleep problems. Inadequate sleep hygiene was

identified as the antecedent variables that appeared to affect sleep. Consequence variables identified to be reinforcing sleep onset delay and night wakings seemed to be breastfeeding, parental attention, comfort that came from co-sleeping, social interaction while awake, and access to food and water. In addition, the mother's presence in bed seemed to act as stimulus control for breastfeeding demands. Upon completion of the assessment and formulation of treatment plan, additional factors were identified during the treatment implementation requiring modifications to the original treatment plan. For example, although breastfeeding was initially identified as reinforcer for night wakings, eliminating night-time breastfeeding did not eliminate night wakings prompting the re-evaluation of breastfeeding as the reinforcer. Ultimately, social attention gained during breastfeeding was identified as the true primary reinforcer. Another change was made to the treatment plan because the child started having a bowel motion immediately upon being put to bed requiring the addition of toileting strategy as part of bedtime routine (McLay et al., 2017). Together with Jin et al. (2013) study, the study by McLay et al. (2017) provided yet another crucial example of the very few studies that have synthesised antecedent and consequence variables together with function-matching assessments to create comprehensive and socially valid treatments for sleep problems in children with ASD based on the individualised outcomes of FBA.

In summary, evidence for effective behaviourally-based treatments on sleep problems in children with ASD is limited. The heterogeneity of ASD symptoms and high rate of comorbidity presents challenges in conducting research in this area. In addition, given that there seemed to be a relationship between sleep problems and ASD symptomatology and that considerable differences in level of functioning and symptom expression were found in children with ASD, certain degrees of treatment individualization are necessary. These modifications provide further challenges on the replicability of treatments and might contribute to the limited evidence of effective treatments (Vriend et al., 2011).

Case Complexity and Treatment Responses

Evidence of positive treatment effects for FBA interventions focusing on multiple components of sleep topography is available in the literature (Friedman & Luiselli, 2008; Jin et al., 2013; McLay et al., 2017; Papadopoulos et al., 2015; Reed et al., 2009; Vriend et al., 2011); however, given the complex array of factors contributing toward sleep problems in children with ASD, further replication of these outcomes is necessary (McLay et al., 2017). The complex array of factors contributing toward sleep problems could also be viewed as factors that can potentially complicate treatment (Delgadillo, Huey, Bennett, & McMillan, 2017). Hence, identification and understanding of this complex array of factors are critical steps in facilitating future replication studies. After all, complex cases require interventions guided by formulations that account for obstacles to improvement (Tarrier, 2006). Understandably, the complex array of factors not only encompass the children (child complexity factors) but also the parents or caregivers (family complexity factors). Common complexity factors had been identified through a survey of practitioners (psychologists and psychiatrists) who worked with children in clinical setting (Kazdin, Siegel, & Bass, 1990, as cited in Kazdin & Whitley, 2006). Common child complexity factors are comorbidity of other disorders commonly observed in children with ASD and the scope and severity of dysfunction. Common family complexity factors are parent dysfunctions (e.g., ASD, depression or other disorders), parental stressors in relationship, socio-economic disadvantage of the family (Kazdin & Whitley, 2006).

Kazdin and Whitley (2006) investigated the relationships between treatment outcomes and comorbidity and complexity of cases in 183 clinically referred children who met criteria for oppositional defiant disorder (also referred to as ODD by some sources) and 132 clinically referred children who met criteria for conduct disorder (also referred to as CD by some sources). In the study, complexity was determined using a number of factors, including scope of symptoms exhibited by the child, academic impairment, history of clinical problem, socioeconomic disadvantage of the family, parent and family functioning, and obstacles in coming to treatment.

Comorbidity was measured by the number of additional disorders diagnosed in the children and number of symptoms that were present across all disorders. These symptoms might capture problems that were clinically significant but might not be apparent enough to trigger a diagnosis. The authors found that children with greater comorbidity showed significantly greater therapeutic change than children with no or only one comorbidity. Children who displayed more symptoms across all diagnoses showed greater therapeutic change. Hence, comorbidity did not diminish the degree of therapeutic change; however, in terms of treatment outcomes, children with greater comorbidity or more symptoms across all diagnoses did not have a different treatment outcome compared to those with fewer comorbidity or symptoms. The authors then evaluated case complexity through four domains: scope and severity of child dysfunction, parent and family functioning, socioeconomic disadvantage, and barriers that emerged during treatment. Scope and severity of child dysfunction included total number of symptoms from the diagnostic interview, history of academic delays and history of antisocial and delinquent behaviour, frequency of delinquent acts in the past year and overall measures of aggression. Parent and family functioning factors included parent dysfunction and psychopathology, parental stressors, and family relationship issues. Socioeconomic disadvantage was included due to its association with many complexities of living, difficulties in managing daily routines and receiving support services. Barriers that emerged during treatments included perceived challenges associated with the treatment, poor relationship with the therapist and perceived irrelevance of the treatment (Kazdin, Holland, & Crowley, 1997). The authors found positive associations between scope and severity of child dysfunction and therapeutic change. More severely dysfunctional children responded better to treatment. An association was only found between overall parent stress and therapeutic change. Children whose parents showed greater stress at pre-treatment displayed greater therapeutic change. No association was found between socioeconomic disadvantage, and therapeutic change, that is, no difference in treatment responses was observed for more disadvantaged cases. Finally, in terms of barriers to treatment, responses to treatments were less for children whose parents experienced greater barriers

in treatment participation. In conclusion, although comorbidity was positively associated with therapeutic change, children with more comorbidities did not exhibit different treatment outcomes at posttreatment compared to other children. Furthermore, excluding barriers associated with treatment participation, other domains of case complexity did not weaken the treatment effects (Kazdin & Whitley, 2006).

Other barriers associated with treatment participation could stem internally from the individual. ASD-related impairments in cognition, communication, and self-regulation could affect the level of participation in various activities and ultimately affect individual's gain from such activities. In one study, Miller et al. (2016) analysed Participation and Activity Limitation Survey (PALS) data on children with neurodevelopmental disorders/disabilities (NDD/D) (ages 5 to 14; weighted $n = 120,700$) in order to investigate the relative roles of an individual's NDD/D related functional characteristics in explaining treatment outcomes. Parental reports were obtained regarding the individual's difficulties in respective functional areas: communication, learning, and limitations due to a psychological, or behavioural, emotional condition. Difficulties with reciprocal social interactions were estimated by using questions on cognitive limitations due to the presence of a developmental disability or disorder, such as ASD, Down syndrome, or mental impairment caused by a lack of oxygen at birth. Results showed that ASD-related functional characteristics (particularly, impairments in cognition, learning, communication, regulation of emotions and behaviour) which explained individual's participation in various activities (Miller, Masse, Shen, Schiariti, & Roxborough, 2013) could possibly lead to the different levels of barriers to treatment. Similarly, another study reported that children's play, social and self-stimulatory behaviours predicted the children's responses to behavioural treatment programme and, ultimately, improvements in the areas of language, play, and social skills (Sherer & Schreibman, 2005). In this study, the authors identified and tested potential predictor variables of children's responsiveness to a widely used behavioural treatment programme – pivotal response training (PRT; Koegel, et al., 1989). The authors analysed archival data of 11 children who received a diagnosis of ASD to

identify specific child characteristics that were present at intake and that might be predictive of treatment outcome. Based on child play behaviours, social behaviours, and self-stimulatory behaviours, two distinct behavioural profiles were formulated. The responder profile was formulated based on the behavioural profiles of children who responded well to PRT. These children tended to exhibit, at the onset, a moderate-to-high interest in toys, had an approachable behaviour, had moderate-to-high rates of verbal stereotypy, and had low-to-moderate rates of nonverbal self-stimulatory. In contrast, the non-responder profile was formulated based on the behavioural profiles of children who did not respond well to PRT. These children showed limited toy play, approach behaviours and verbal self-stimulatory behaviours. In addition, these children exhibited moderate levels of avoidant behaviour and nonverbal self-stimulatory behaviour at intake. The responder and non-responder behavioural profiles were used to select and then predict treatment outcomes on six participants – three who matched the responder profile and three who matched the non-responder profile. As predicted, participants with the responder profile exhibited improvements in the areas of language, play, and social skills. Participants with the non-responder profile did not show any improvement. Given that PRT was play based and focused on areas of the child's developments including motivation, response to multiple cues, initiation of social interactions, and self-management (Autism Speaks Incorporated, 2018), the authors showed that behavioural profiles formulated based on specific behaviours – functional play, approach behaviours, avoidant behaviours and self-stimulatory behaviours – could be used as a predictor for treatment outcomes. Nevertheless, the authors noted that, based on parent reports, children with a non-responder profile in the study responded well to intensive home-based discrete trial training (DTT; Lovaas, 1987). Hence, the non-responder profiles proposed in the study was specific to PRT only and might not indicate the general responsiveness to other treatment programmes. Interestingly, in an earlier study by Koegel et al. (2003), non-responder children with ASD demonstrated a favourable response to treatment after completing training to self-initiate (Koegel, Carter, & Koegel, 2003).

Building on the responder profile identified in the Sherer and Schreibman (2005) study, Fossum et al. (2018) investigated the generality of the responder profile and incorporated child affect as a potentially important variable. The authors reported that better baseline expressive language and cognitive abilities, more positive affect and appropriate toy contact, and lower levels of social avoidance and stereotyped/repetitive vocalisations were the characteristics of children who gained the most from the treatment (Fossum, Williams, Garon, Bryson, & Smith, 2018). The recent Fossum et al. study (2018) supported the behavioural features of children who responded to the PRT treatment despite the differences in terms of age range and level of cognitive functioning and verbal ability that study and the original Sherer and Schreibman (2005) study. Most importantly, the inclusion of positive affect in the responder behavioural profile in the recent study pointed to the possibility that positive affect served as an indicator of the child's motivation for learning (Koegel et al., 1998; as cited in Fossum, 2014) that might facilitate the therapist's work by providing an indication of what activities or rewards can be used to greatest advantage for promoting communication (Fossum et al., 2018).

In conclusion, this study intends to contribute to the identification and understanding of case complexity in treatment formulation and selection for sleep problems in children with ASD. In the following chapter, a literature review will be performed to obtain conceptual clarity and literature evidence surrounding this topic.

Chapter 2

Literature Review

The purpose of this literature review is to explore child-specific complexity factors that may serve as predictor variables for treatment responses. Although in the context of child therapy, parent- and family-specific complexity factors are a significant part of the equation, the focus of this study will be on the child-specific factors. A separate and concurrent study, that does not form part of this thesis, examines the parent- and family-specific factors.

In the present study, the hypothesis that is evaluated is that the complexity of a case influences the effectiveness of an evidence-based treatment for sleep problems in children with ASD and that the greater the child complexity the less responsive children may be to a behavioural sleep treatment programme. There are no standardised ways of assessing case complexity and complexity can include an almost infinite set of factors. Despite this many of the factors could be identified through an examination of the limited existing research, parental or caregiver reports and from clinical processes. These case-specific complexity factors include the scope and severity of dysfunction, severity of sleep problems, comorbidity with other disorders, and medications taken to address ASD and other symptoms and is largely based upon those factors identified by Kazdin and Whitley (2006).

Search Process

A systematic search was conducted to identify relevant studies investigating associations between biological, developmental, and psychopathological characteristics and outcome of treatments for sleep problems in children with ASD. The following databases were included in the search: PsycINFO, PsycARTICLES, ScienceDirect, PubMed/MEDLINE (Ovid), and ProQuest. In addition, the reference lists of obtained articles, systematic reviews, and meta-analyses were read to identify any additional articles that were relevant to the review and not found in the above

databases. Searches were limited to those articles that were written in the English language, published in a peer-reviewed journal, and that included participants 18 years of age or under.

The search strategy addressed key search terms for sleep issues, characteristics of complexity and treatment outcomes. More specifically, searches were conducted using a combination of the following key terms: sleep (disturbance, problems, difficulties, phenotype), diagnostic (Autism Spectrum Disorder, autism), characteristics (characteristics, complexity, phenotype, subgroup, dimension), and treatment outcome keywords: response, treatment response, outcome, treatment outcome. Age-related key terms such as child were not included in the search phase to avoid missing studies that did not include age-related terms in the title of the study or that used specific terms (e.g., pre-schoolers). Titles and abstracts were screened for relevance and content.

Children and Adolescent Characteristics and Treatment Responses

Whilst there is strong evidence linking sleep problems and the child comorbidity and complexity (i.e. scope and severity of ASD) in children with ASD, limited research has been conducted to investigate the impact that these variables have on children's response to interventions, including those that are targeting sleep problems. Several studies have investigated the association between child characteristics and the outcome of behavioural intervention in general. For example, multiple studies have been conducted to investigate the association between individual characteristics and responses to early intensive behavioural interventions (EIBI). In many cases, it was found that treatment response was related to child characteristics as opposed to the amount or type of intervention (Eaves & Ho, 2004).

Strauss et al. (2013) performed a comprehensive synthesis of six meta-analyses of EIBI for children with ASD published from 2009 to 2011 (Eldevik et al., 2009; Makrygianni & Reed, 2010; Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011; Reichow & Wolery, 2009; Virues-Ortega, 2010). All treatment programmes in the meta-analyses were based on applied behaviour analysis with varying definitions – from EIBI based strictly on the Lovaas (1987) method to broader

definitions of comprehensive treatment programmes including Pivotal Response Treatment and Group Intensive Family Training. Several variables were analysed to identify whether they related to treatment outcome. This included pre-intervention characteristics (e.g., IQ, language skills, adaptive functioning, severity of ASD symptomatology), chronological age, gender, increase in IQ or skill acquisition during treatment, and treatment characteristics (intensity and duration of treatment, treatment supervision, supplement treatment and parent involvement). The results of this meta-analysis were mixed. Magiati et al. (2007) found that pre-intervention characteristics (i.e. initial IQ combined with language skills) was the strongest predictor of treatment outcome. However, two studies found that pre-intervention characteristics (i.e. initial IQ alone) does not best predict outcome but increase in IQ scores during treatment progress predicted outcomes (Ben-Itzhak, Lahat, Burgin, & Zachor, 2008; Sallows & Graupner, 2005)

More recent studies across various EIBI models continued to show possible associations between treatment responses and pre-treatment factors such as symptom severity, imitation, and functional use of objects (Vivanti et al., 2013), adaptive skills (Flanagan, Perry, & Freeman, 2012), and play skills (Kasari, Gulsrud, Freeman, Paparella, & Hellemann, 2012). Importantly, Eapen et al. (2013) suggested that the genetic and phenotypic heterogeneity inherent in ASD might prevent a single EIBI treatment from being universally effective and many nuanced treatment approaches may be required (Eapen et al., 2013).

Age at baseline and treatment responses. From a behavioural and developmental theory perspective, it could be argued that starting Intensive Behavioural Intervention (IBI) at a younger age might be beneficial (Perry et al., 2011). It had been suggested that a period of development exists before the age of 4 during which early interventions aimed at stimulating young children at risk for ASD can substantially change the course of both brain and behavioural development (see Dawson, 2008). Studies that have examined response to treatment in children across age-ranges have typically found better treatment outcomes for younger children when compared to older children (Flanagan et al., 2012; Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009; Harris &

Handleman, 2000; Mandell et al., 2013; Perry et al., 2011). For example, in a community-based study for children with autism, aged 3.5- to 4.5-year-old, comparing 61 children who received IBI and 61 individually matched children from a waitlist comparison group, Flanagan et al. (2012) reported that initial age is an important predictor of better outcomes in IBI relative to the comparison group. In another community-based study for children with autism with a wider age range (aged 2-7 years), Perry et al. (2011) reported that children who were most successful in the program were under 4 years of age at program entry. Using a sample of older children, aged 5- to 8-year-old, Mandell et al. (2013) investigated the efficacy of a classroom-based intervention for children with ASD in a randomized field trial of 119 students and reported that each additional year of student age was associated with a decrease in treatment outcome. In contrast, in a study of a similar classroom-based intervention for children with ASD using a sample of 191 children aged 5- to 8-year-old, Pellecchia et al. (2015) reported that increased child age was not associated with differences in outcome. However, given the data used in the Pellecchia's (2015) study were selected from the third year of a larger intervention study, the authors suggested that involvement in the intervention study during the previous years might have affected the association between student age and outcome because (1) skills of teaching older children with autism might have improved, (2) older children might have reached their maximum response to interventions if they had been receiving intensive intervention for several years, and (3) there were more higher functioning older children in the third year of the intervention than in previous years. Other studies also reported no association between initial age and treatment outcome (Eikeseth, Smith, Jahr, & Eldevik, 2007; Makrygianni & Reed, 2010; Vivanti et al., 2013). Nevertheless, with the exception to the Pellecchia's (2015) study, all the other studies had a small sample size ($n < 30$). Clear association might not be detected due to several reasons, including specific selection criteria, a relatively narrow age range at intake, and a small sample size (Flanagan et al., 2012; Perry et al., 2011). In summary, the precise nature and power of age as a predictor for IBI treatment outcomes remained less obvious (Matson & Smith, 2008). Similarly, given that sleep problems of children with ASD

were more likely to persist as they aged and sleep problems were reported to peak around the age of 6 to 9 years old (Hodge et al., 2014; Krakowiak et al., 2008), it can be reasonable to assume different responses to treatment would exist depending on the age of the treatment group.

Adaptive behaviour at baseline and treatment responses. For children with ASD, some critical adaptive behaviours include the capacity for conversing with and understanding others, interacting with others, developing relationships, and participating in group activities (Klin et al., 2007). Studies had suggested distinct sleep patterns based on the developmental trajectories of adaptive behaviours in children with ASD (Anderson, Oti, Lord, & Welch, 2009; Baghdadli et al., 2012; Cohen et al., 2017). Anderson et al. (2009) conducted a longitudinal study of 93 children with ASD to examine prospectively the development of adaptive social skills between ages 2 to 13. All children received parent-implemented behavioural therapy (e.g., speech therapy, ABA, parent-implemented structured teaching). Two distinct growth rates of adaptive skills in the domains of social and communicative skills were found over time. One group of children showed dramatic increases in social skills over time, with VABS scores improving to near age norms for typical development by the age of 13 – an average increase of about 8 years, 11 months over the 11-year period. This group of children exhibited stronger expressive but not receptive language skills at age 2. The second group of children showed a much more modest increase in adaptive social skills – with an increase of 21 months over the 11-year period. In another longitudinal study of 152 children with ASD, aged 3.6- to 6-year-old, over a 10 year period, Baghdadli et al. (2012) reported two different growth trajectories in the domains of socialization and communication for high functioning and lower functioning children. High functioning children tended to have better expressive language and less severe autistic symptoms during childhood. In a more recent study using 50,000 nights of care-giver sleep/wake logs collected from 106 children with low functioning autism, aged 5 to 18 years old, Cohen et al. (2017) also identified two distinct sleep phenotypes with characteristics differences in the severity of key sleep features – total sleep time, sleep efficiency, frequency of night waking, and sleep onset. The group with highly variable and unstable

sleep had significantly reduced full-scale IQ scores, communication scores, daily living, and overall adaptive behaviour composite scores, as measured by the VABS. No significant differences between the two distinct sleep phenotype groups in terms of age, gender, psychiatric comorbidities, medical comorbidities and use of medications that could affect sleep. The identification of these distinctive sleep profiles with robust association with adaptive functioning in children with low functioning autism would enable more individualized and effective therapies to be formulated and implemented. However, adaptive behaviour skills at baseline were seldom examined as predictors of treatment outcome; instead adaptive behaviour skills were often examined post-treatment and included as outcome measures (Perry et al., 2011). Studies consistently reported that pre-treatment higher adaptive behaviour skills, as measured using the VABS, were associated with positive response to treatment across various EIBI models (Flanagan et al., 2012; Perry et al., 2011; Remington et al., 2007; Sallows & Graupner, 2005). Furthermore, initial adaptive behaviour skills predicted better performance in children receiving treatments other than EIBI (Eaves & Ho, 2004; Eikeseth et al., 2007; Flanagan et al., 2015; Kim, Macari, Koller, & Chawarska, 2016; Magiati, Moss, Charman, & Howlin, 2011).

IQ and Cognitive ability at baseline and treatment responses. It had been established that lower-functioning individuals have more severe symptoms and poorer coping skills and therefore exhibit more sleep problems than higher-functioning individuals (Hollway et al., 2013). Also, a relationship between sleep, maturational cognitive processes and adaptive skills has been established (Dan & Boyd, 2006). Hence, level of intellectual functioning (i.e. IQ and cognitive ability) at baseline would likely to provide some indications regarding the severity of sleep problems and, consequently, the level of sleep treatment responses. In children with ASD, Hollway et al. (2013) reported a positive association between IQ and Sleep Anxiety scores on the CSHQ, although IQ did not predict other sleep outcomes such as CSHQ Sleep Duration, CSHQ Bedtime Resistance, and CSHQ 23-item Total Scores.

The impact of cognitive ability on treatment outcome has been examined in several studies (Ben-Itzhak & Zachor, 2007; 2011; Ben-Itzhak, Watson, & Zachor, 2014; Eaves & Ho, 2004; Eldevik, Hastings, Jahr, & Hughes, 2012; Hayward, Eikeseth, Gale, & Morgan, 2009; Thurm, Lord, Lee, & Newschaffer, 2007; Turner & Stone, 2007) and is widely reported as a predictor of response to behavioural treatment in children with ASD. (i.e., higher cognitive ability at baseline is a strong predictor of improvements in cognitive ability, adaptive behaviour, language skills and ASD diagnostic classification following treatment) (Ben-Itzhak et al., 2014; Bibby, Eikeseth, Martin, Mudford, & Reeves, 2002; Eikeseth, Smith, Jahr, & Eldevik, 2002; 2007; Eldevik, Eikeseth, Jahr, & Smith, 2006; Hayward et al., 2009; Lovaas, 1987; Smith, Groen, & Wynn, 2000; Turner & Stone, 2007; Zachor, Ben-Itzhak, Rabinovich, & Lahat, 2007). Similarly, low IQ and absence of language at 36 months predicted limited progress (Sallows and Graupner, 2005).

Scope and severity of autism symptoms and treatment responses. Severity of autism symptoms can be defined based on the diagnosis of autism subtypes or levels of autism severity (Matson & Smith, 2008; Remington et al., 2007; Sallows & Graupner, 2005). However, a variety of measures of autism severity was used in past studies on children with ASD making comparison difficult (Howlin, Magiati, & Charman, 2009). Assuming more severe symptoms of ASD would result in poorer outcome is a logical first step in investigating the role that severity of autism symptoms has on treatment response. However, severity of autism symptoms has rarely been included as a potential predictor of outcome (Matson & Smith, 2008). Research findings were mixed. Some studies showed milder or lower baseline autism scores were predictive of better treatment responses (Eaves & Ho, 2004; Sallows & Graupner, 2005). In a 4-year longitudinal study of twenty-four children, aged 2 years 9 months to 2 years 10 months, with ASD receiving early intensive behavioural treatment, Sallows & Graupner (2005) reported that lower baseline autism scores, as measured by Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & LeCouteur, 1994) scores (together with higher IQ and more rapid early skill mastery) were predictive of rapid treatment responses. Based on a sample of forty-nine 2 year olds with social and language

characteristics suggestive of autism, Eaves and Ho (2004) also reported that higher functioning children with milder autism, based on the Childhood Autism Rating scale (Schopler, Reichler, & Renner, 1988), were the most improved. In a separate study of predictors of treatment outcomes for children, aged 1 year 10 months to 4 years 10 months, with ASD receiving Early Start Denver Model (ESDM; Rogers & Dawson, 2010) in a group setting, Vivanti et al. (2013) reported that less severe autism symptoms at baseline, as measured by Autism Diagnostic Observation Schedule severity score (ADOS; Lord, Rutter, DiLavore, & Risi, 1999), appeared to play a role in predicting treatment gains in both expressive and receptive language. Ben-Itzhak & Zachor (2007) conducted a study of 25 children, aged 1 year 8 months to 2 years 8 months, with ASD. The study focused on severity of autism symptoms in the area of language and communication and reciprocal-social interaction, as measured by the ADOS. Using median scores in the language and communication section, children were divided into high and low communication groups. Similarly, using median scores in the reciprocal-social interaction section, children were divided into high and low social groups. Children with higher initial cognitive levels and fewer early social interaction deficits showed better acquisition of receptive language, expressive language, and play skills, although, severity of the measured communication deficits was not related to outcome in any developmental domain. In a more recent study with a very large and diverse sample of children ($n = 332$), aged approximately 2- to 7-years-old, Perry et al. (2011) reported that children with milder or fewer autism symptoms at intake, as measured by the Childhood Autism Rating (CARS; Schopler, Reichler, & Renner, 1988) scale, were more likely to achieve positive responses to treatment. In contrast, a randomized control trial was conducted with two groups of pre-schoolers, aged between $2\frac{1}{2}$ years and $3\frac{1}{2}$ years, with ASD. There were twenty-three pre-schoolers in the treatment group and twenty-one pre-schoolers in the comparison group. Assessments were taken prior to intervention, after 1 year and after 2 years of intervention. More autistic symptoms of autism at baseline, as reported on the Developmental Behaviour Checklist - Autism Screening Algorithm

(Einfeld & Tonge, 2002) by both parents, were associated with more positive responses to treatment (Remington et al., 2007).

Gender differences and treatment responses. The presentation of ASD clinical symptoms are different between male and female (Lai, Lombardo, Auyeung, Chakrabarti, & Baron-Cohen, 2015) and seemed to be influenced by age. Studies of children, aged 1 year 4 months to 3 years 10 months, concluded that there were no major sex differences in ASD manifestation. However, sleep problems and anxious or depressed affect were more prevalent in girls (Hartley & Sikora, 2009; Reinhardt, Wetherby, Schatschneider, & Lord, 2015). In contrast, studies of older children and adolescents with ASD, aged 3 to 18 years, found gender differences in restricted interests, hyperactivity, and social anxiety. Higher hyperactivity levels were common in males and social anxiety were more prevalent in females (May, Cornish, & Rinehart, 2014). Females with ASD who were cognitively abled had substantially lower levels of restricted interests and those with lower cognitive ability had greater social communication impairment, weaker adaptive skills and greater externalizing problems relative to males (Frazier, Georgiades, Bishop, & Hardan, 2014; Frazier & Hardan, 2017; Mandy et al., 2012). However, gender is rarely considered when making diagnostic or treatment decisions (Pinares-Garcia, Stratikopoulos, Zagato, Loke, & Lee, 2018). Hence, major advancements in the prevention or treatment of ASD in both males and females can be achieved by a better understanding of the gender difference (Halladay et al., 2015). In a recent study of thirty-five 3-year-old children with ASD, Tiura et al. (2017) included gender in the study to control for any gender differences that might arise in parent reported outcome measures. Results showed that male participants tended to improve more quickly in the areas of adaptive behaviour and physical development; however, the authors noted that the small sample of female participants might have resulted in limited variability, thus affecting the result. In contrast, in a longitudinal study of patterns of growth in verbal abilities among 2-year-old children with ASD assessed at age 3, 5 and then 9 years old, Anderson et al. (2007) found that gender was not predictive of treatment outcomes.

Externalising/Internalising behaviour problems at baseline and treatment responses. In typically developing children, sleep problems are associated with behavioural difficulties. However, the directionality of this association is unclear. Furthermore, the question of whether different behavioural difficulties are associated with different type of sleep problems remains to be answered. Understanding of these associations will undoubtedly improve treatment responses (Quach, Nguyen, Williams, & Sciberras, 2018). In a longitudinal study of 4983 typically developing children aged 4 to 5 years old, Quach et al. (2018) reported that interventions targeting externalising, but not internalising, difficulties might improve childhood sleep and further research were necessary to investigate whether sleep problem interventions decreased the occurrence of both externalising and internalising difficulties. Sleep problems are also associated with behavioural difficulties in children with ASD (Goldman et al., 2009; 2011; Malow et al., 2006a; Mayes & Calhoun, 2009). In a longitudinal study of 437 children, aged 2 to 10 years, with ASD assessed at baseline and follow-up 3.8 years later, sleep problems at baseline predicted somatic complaints in older children. Negative longitudinal effects of sleep disturbance were found on attention and hyperactivity in children with ASD. However, no significant predictive relationship was found between sleep problems and anxiety. Nevertheless, treating sleep problems might have a positive impact on attention and behavioural regulation and future research is needed to determine whether treatment of sleep problems prevents or reduces inattention and hyperactivity in early childhood for children with ASD (Mazurek, Dovgan, Neumeyer, & Malow, 2019).

Studies on the association between problem behaviours and responses to early intensive behavioural intervention could provide some insights relevant to the study of problem behaviours and responses to treatment for sleep disturbances. In a two-year longitudinal study of early intensive behavioural intervention for children with ASD, Remington et al. (2007) reported that more problem behaviours, as measured by the Developmental Behaviour Checklist (DBC; Einfeld & Tonge, 2002), were associated with more positive responses to treatment. There was, however, no reduction in parental reports of children's behaviour problems, following early intensive

behavioural intervention, which came as a surprise to the authors and was attributed to the absence of detailed functional analysis and function-informed interventions for problem behaviours were not the prominent components. Nevertheless, positive benefits of behavioural intervention had been expected given the known association between behaviour problems and severity of cognitive and adaptive functioning, especially in language or communication skills (Remington, et al., 2007). In another study, Henderson et al. (2016) reported that, in comparison to non-autism group ($n = 57$), parents of children with ASD ($n = 58$) reported higher rates of externalising behaviours and significantly less frequent general routine (i.e., daily living routines, household responsibilities, homework routine, and discipline routine). No interaction was found between diagnostic group and consistency of bedtime routines in predicting externalising behaviour problem and the authors reasoned that externalising behaviours might be influenced by multiple factors in addition to bedtime routines (Henderson, Barry, Bader, & Jordan, 2011).

In one of the first studies to explore observational relations between sleep and behaviour in the context of intensive behavioural intervention, Abel et al. (2018) conducted a study using a sample of 39 children, aged 2 to 10 years, with ASD receiving intensive behavioural intervention. Results showed no significant day-night associations between children's sleep and challenging behaviours. Instead, average patterns of sleep over time appeared to be more influential in challenging behaviours than daily fluctuations in sleep. Despite receiving intensive behavioural intervention, children who slept less (on average) exhibited significantly higher rates of repetitive behaviour. Also, children who had more night wakings (on average) during the week engaged in higher rates of negative affect (Abel, Schwichtenberg, Brodhead, & Christ, 2018).

Co-morbidity and treatment responses. Additional psychopathology or conditions were likely to exacerbate problems associated with treatment effectiveness (Matson & Smith, 2008). Studies pointed to the associations of co-occurring presence of psychological symptoms and treatment responses. For example, in a study of 152 students with ASD after one year of school-based behavioural intervention (discrete trial training, pivotal response training, and teaching within

functional routines), Pellecchia et al. (2016) examined associations between treatment outcome, as measured by changes in cognitive ability, and child characteristics (including age, social skills, adaptive behaviour, autism severity, autism core symptoms, language ability, and co-occurring presence of psychological symptoms). The study found that age and co-occurring presence of psychological symptoms, specifically the presence of symptoms associated with social anxiety, were associated with differences in treatment outcome. Although the study recommended that further examination on the presence of symptoms associated with social anxiety to identify the individual component of the construct of social anxiety (either social fearfulness or social motivation) and to include treatments for addressing the particular component of social anxiety, the important implication presented by the Pellecchia et al. (2016) study was that, given the association between increased social anxiety symptoms and poorer treatment outcomes, addressing the co-occurring presence of psychological symptoms early might have a profound impact in maximising treatment outcomes.

Medications and treatment responses. Children with ASD are often prescribed medications to treat autism symptoms or comorbid psychopathology, including depressions, ADHD, irritability and so on; however, some of these medications might have effects on sleep and, hence, might improve or worsen sleep problems. For example, antidepressants with sedative properties (e.g., Mirtazapine, Doxepin, Trazodone) rapidly improve sleep, but might cause problems in long-term treatment due to over-sedation. Others with activating effects (e.g., Venlafaxine, Fluoxetine) might disrupt sleep at least in short-term treatment. In addition, some antidepressants might also worsen or induce sleep disorders like sleep bruxism, restless leg syndrome, rapid eye movement sleep behaviour disorder, sleep apnoea, and nightmares (Wichniak, Wierzbicka, Walecka, & Jernajczyk, 2017). Studies have also shown that children with ASD prescribed with psychostimulant medications (Methylphenidate and Atomoxetine) considered to be effective to treat ADHD-like symptoms in autism (Clemow, Bushe, Mancini, Ossipov, & Upadhyaya, 2017; Cortese, Castelnau, Morcillo, Roux, & Bonnet-Brilhault, 2012) reported adverse

effects of irritability, insomnia and emotional outbursts associated with taking Methylphenidate (Posey et al., 2007; Simonoff et al., 2013); and fatigue, early morning waking, and initial insomnia due to Atomoxetine (Harfterkamp et al., 2012). Similarly, in large controlled psychopharmacology trials in children with autism administered with Risperidone or Aripiprazole (medications for treatment of irritability associated with ASD), common adverse effects reported were fatigue, sedation and drowsiness (Marcus et al., 2009; McCracken et al., 2002). Clearly, any medications that had effects on sleep might influence the implementation, efficacy and success of behavioural based interventions.

In summary, for initial age, adaptive skills, severity of autism, gender, and externalising or internalising behaviours, results are not completely consistent and further research is needed (Henderson et al., 2011; Mazurek et al., 2019; Pellecchia et al., 2016; Perry et al., 2011; Remington, et al., 2007; Tiura et al., 2017). In contrast, higher IQ and cognitive ability has more consistently predicted treatment responses (Ben-Itzhak, Watson, & Zachor, 2014; Fossum et al., 2018; Howlin et al., 2009; Reichow, 2012) and medications prescribed to children with ASD might have effects on their sleep (Harfterkamp et al., 2012; Marcus et al., 2009; McCracken et al., 2002; Posey et al., 2007; Simonoff et al., 2013; Wichniak et al., 2017).

Limitations of Existing Research

Overall, knowledge of child characteristics that were associated with differences in treatment responses is limited (Sherer & Schreibman, 2005). The majority of previous studies had investigated child characteristics that were predictive of treatment responses in intensive behaviour intervention and very limited studies were conducted to investigate child characteristics as a predictor of response to treatment of sleep disturbance. Nevertheless, relying on existing research on intensive behaviour intervention to inform the choice of individual-specific factors to investigate in the present study seemed to be a reasonable starting point.

Unfortunately, many of the previous studies that have investigated the relationship between case complexity and treatment responses are subject to a number of limitations.

First, in most cases, intervention type, intensity and duration might have varied resulting in non-standardized and individualized interventions employed in the sample potentially confounding the observed findings and preventing analysis of which factors contributed to the outcomes (Knight & Johnson, 2014; McLay, France, Knight, Blampied, & Hastie, 2019; Pellecchia et al., 2015).

Second, limited numbers of participants were identified for specific behavioural interventions. Single-case study design was often used and no control group was included in the majority of the studies. Although as an important first step to begin testing an intervention, not including a control group was acceptable since the use of baseline period allowed for comparison of behaviour before and during treatments and any improvement observed during the intervention period relative to baseline could be viewed as a benefit of the intervention (Smith et al., 2007). However, the limited number of participants in many of the studies causing the studies to be underpowered to study predictors of outcome (Pellecchia et al., 2015).

Third, child characteristics (i.e., cognitive ability, autism symptom severity, adaptive behaviours, language skills) investigated as predictors of treatment outcome were not directly modifiable because these constructs reflect a variety of distinct underlying processes, making it difficult to understand the specific mechanisms underlying treatment response (Fossum et al., 2018; Vivanti, Prior, Williams, and Dissanayake, 2014).

Fourth, previous studies did not account for the presence of co-occurring psychological symptoms or concurrent medications, and their potential effects on the responses to treatments in children with autism (Masi et al., 2017; Pellecchia et al., 2016).

Fifth, reported findings might not be generalized to the larger population of children with ASD because sample used in the study was children referred to outpatient or university-based clinics rather than children selected from community-based settings (Pellecchia et al., 2016).

Sixth, the role of co-variates such as age, socio-economic status, and family factors were not evaluated to determine the impact of these factors had on the responses to behavioural sleep interventions (Rigney et al., 2018).

Seventh, limited randomized controlled trials were performed. Well-powered clinical trials would enable estimation of potential benefits of a specific intervention and exploration of additional factors, such as identification of specific individuals who would benefit most from the intervention and the reasons behind such benefits (Vivanti et al., 2018). A randomized controlled trial would determine the effectiveness of the intervention and strengthen external validity of the findings (Sanberg, Kuhn, & Kennedy, 2018).

Rationale

This review has demonstrated the relationship between sleep problems and children's autism symptoms and severity (e.g., social interactions and communication difficulties, stereotyped and repetitive behaviour), adaptive behaviour, challenging behaviour (externalising and internalising), comorbid symptoms (e.g., hyperactivity and inattention). In addition, the relationship between case complexity (e.g., biological, psychological, behavioural, prescribed medications) and treatment responses has been explored. The child characteristics that were predictive of treatment responses in other behavioural interventions served to inform the choice of individual-specific factors to investigate in the present study.

Hence, the present study adds to the literature by examining the extent to which biological, developmental, and psychopathological characteristics predicted treatment outcome for sleep problems in children with ASD. By focusing only on individuals with an ASD diagnosis and not comparing with typically developing individuals, the understanding of any individual and subgroup differences within the autism spectrum, and how these differences affected treatment responses would allow for individual customisation of treatment programmes and for improving the effectiveness of such programmes (Georgiades, Szatmari, & Boyle, 2013). Furthermore, by identifying these differences and understanding the impact that these differences have on treatment responses, there will be opportunities to individually tailor and increase the effectiveness of treatment programmes (Eapen et al., 2013). Specifically, this study investigates the following research questions:

(1) Did the treatment alter the child's sleep to a clinically significant degree?

(2) Is there a latent complexity variable affecting the child's responses to treatment?

(3) Do the latent complexity variable and any children's characteristics (i.e., gender, age, autism core symptoms severity, co-morbid psychopathology, sleep problems severity, behaviour severity, and medications) serve as predictors of treatment responses?

By answering these research questions, efforts will be made to identify critical factors so that these factors can be addressed early to maximise treatment outcomes.

Chapter 3

Method

This thesis involved the examination and analysis of pre- and post-treatment psychometric assessment data collected from a subset of children with ASD who were enrolled in a larger study evaluating the effect of behavioural interventions for sleep problems. This chapter describes the current project within the context of the wider sleep and autism research study being undertaken by a team of researchers at the University of Canterbury.

The present study in the context of the larger sleep and autism study

The larger study focused on investigating the effectiveness of behavioural interventions for sleep problems in children with ASD. In this study, individualised interventions were formulated based on the outcome of Functional Behaviour Assessment (FBA). The effect of these interventions was then evaluated. The focus of the present study was to examine the relationship between child complexity variables and treatment response in children with ASD who participated in this larger study. A post hoc analysis of variables was conducted in order to identify predictors of treatment responses.

The Sleep Research Team

The Sleep Research Team at the University of Canterbury is comprised of two senior academics, registered intern psychologists, PhD and Masters thesis students.

Ethics and Participant Consent

Ethical approval was obtained from the University of Canterbury Human Ethics Committee as a part of the larger Sleep and Autism Study (HEC #2014/150). This study drew upon archived data from the larger study and no additional approvals or consents were required.

All parents were required to provide consent for their child to participate in the Sleep and Autism study. An initial phone discussion was carried out with each family to ensure caregivers or

parents fully understood the nature of the study. Children were informed verbally regarding the study and its requirements. A copy of the information sheets and consent forms were then provided to children/young people and their parents (see Appendix A to D). Before the start of the study, parents/caregivers and children provided informed consent or assent, respectively.

Research Design

For the larger Sleep and Autism study, a non-concurrent single-case multiple baseline across participants design was used to examine the effects of function-based behavioural interventions. As each case was unique, the corresponding treatment plan was individualised. Use of a single case research design allowed for close monitoring of the intervention effects and opportunities to refine the intervention techniques. Single case research design is also a useful starting point for establishing efficacy because such designs provide evidence that the intervention technique has a clear and replicable effect on a specific behaviour (Smith et al., 2007).

Study Phases in the larger Sleep and Autism study

The Sleep and Autism study consisted of the following phases.

Assessment. All questionnaires analysed in the present study were administered as part of assessment conducted during the larger Sleep and Autism study. In addition to the administering the measures, FBA was conducted. Information gathered was used for treatment formulation, planning, and measuring changes following intervention. During the assessment phase, parents were advised to respond in the usual manner and not making any changes to the child's sleep patterns.

Clinical interview. During the assessment phase of the larger Sleep and Autism study, an open-ended clinical interview was conducted by a registered psychologist or a registered intern psychologist who was under supervision of a clinical psychologist. To guide the functional analysis of the child's sleep problems, the Sleep Assessment Treatment Tool (SATT; Jin et al., 2013) was used. The SATT includes questions that investigate and identify (1) the history of a child's sleep problems; (2) the parent or caregivers goals for intervention; (3) the nature and severity of the presenting sleep problems (bedtime routine issues, delayed sleep onset, sleep interfering behaviour,

night wakings, early morning wakings); (4) antecedent and consequence variables that precipitate or maintain the sleep problem; (5) the current pre-sleep routine and sleep schedule; (6) the child's sleep setting; (7) any sleep dependencies (i.e., events lead to bedtime, items needed during bedtime); and (8) any sleep interfering behaviour. During the interview, information was also sought in regard to (1) any previous attempts to address the sleep problem; (2) possible risk factors (i.e., parental psychopathology, drugs or alcohol issues, marital discord); (3) past or present medical or physical conditions that may have an impact on the child's sleep (i.e., comorbid psychopathology, asthma, obstructive sleep apnoea).

Baseline. Each participant was randomly assigned a baseline length of one, two, or three weeks. During the baseline phase, parents were advised not to modify their responses to the child's behaviour and not change the sleep routine. Parents would collect sleep diary data and video recordings. Both the information obtained from clinical interview and sleep data obtained during the baseline phase was used to inform the intervention plan.

Intervention. Intervention commenced immediately upon conclusion of baseline. Parents continued to record sleep diary data and video recordings throughout the intervention phase. Sleep interventions for children were individualized based on the outcome of the FBA, and typically consisted of antecedent and consequence-based modifications (e.g., modifications to sleep hygiene and bedtime routines, modification to sleep/wake schedules, use of visual supports (e.g., Gro Clocks), social stories, stimulus substitutions, faded parental presence, extinction, and reinforcement procedures.

In each case, the goal of treatment was to promote sleep-conducive behaviour and to eliminate sleep interfering behaviours. and the treatment plan was developed in collaboration with the parents (i.e., it integrated their goals and preferences). Parents or caregivers maintained regular contact with researcher throughout treatment via the phone, email or SMS text. Regular contact was maintained to provide motivation, increase parents' confidence in implementing the treatment plan, and resolve issues with expediency. Treatment was maintained until notable progress had been

made, as agreed with the parents or caregivers, until the sleep problem had been resolved, or until the parents chose to withdraw from the study.

Maintenance. The maintenance phase commenced immediately on completion of treatment. During this phase the researcher did not initiate any contact with the child's parents. During this phase, parents and children would have the opportunity to maintain the newly acquired behaviours in their daily lives (Blampied, 2013). At the conclusion of this phase, a semi-structured interview was conducted with parents or caregivers to gain feedback on their level of satisfaction with the treatment outcomes and post-treatment questionnaires were administered.

Follow-up. Short- and long-term follow-up data was collected 4-6 and 10-12 weeks post-treatment respectively. During this phase, parents or caregivers collected one week of sleep diary data and video recordings. The purpose of the follow-up phase was to assess the maintenance of treatment effects over time.

Present study

For the present study, a retrospective pretest-posttest research design was used to investigate the predictors of response to function-based interventions to treat sleep problems among children with a diagnosis of ASD. Cases were drawn from those who completed treatment and provided information for at least one, follow-up phase. Random assignment to treatment and control groups were difficult to carry out for practical and/or ethical reasons. All children in the larger Sleep and Autism study were referred to the study due to the seriousness of their sleep disturbances. Hence, it was not possible to randomly assign children to the treatment program or to no treatment program. Two important steps were taken to determine that it was the treatment that brought about change for each child. First, a multiple-baseline was used in the larger Sleep and Autism study to confirm a replicated pattern of change from baseline to intervention supported by relatively little delay between implementation of treatment and change (Barlow, Nock, & Hersen, 2009). Second, a large clinically significant treatment effect should be observed in the SPS scores at either short- or long-term follow-up, as evidence that the combination of non-specific (Barker, Pistrang, & Elliot, 2013)

and specific treatment effects had been highly positive for these children and their families (McLay et al., 2019).

Participants in the larger Sleep and Autism study

Recruitment. For the larger study, participants were recruited from organizations throughout New Zealand that provide services to children with ASD, via the networks of the research team, or through self-referral.

Inclusion/exclusion criteria. Participants were enrolled in the larger study if they met the following inclusion criteria: (1) aged between 3 and 18 years of age; (2) had a formal diagnosis of ASD made by a paediatrician, registered psychologist or psychiatrist; (3) had parent-reported sleep issues including, but not limited to, delayed sleep onset (time taken to fall asleep), frequent and prolonged night wakings, early morning awakenings, and/or co-sleeping. Children with a medical condition that may affect their ability to follow the study procedures were excluded from the study. Data from participants in the Sleep and Autism study who completed the treatment phase was examined and analysed.

Recruitment for the present study. Participants were 31 (of 42) children who had participated in the Sleep and Autism study who also met criteria for the current study.

Participants in the present study. Data were obtained from 6 girls and 25 boys between 3 and 14 years of age. All children had a formal diagnosis of ASD. Rather than drawing a comparison with typically developing children, the present study aimed to understand the meaning of individual and subgroup differences on treatment outcomes within the children with a diagnosis of ASD. A summary of participant characteristics is presented in Table 1.

Measures

Sleep measures.

The following measures were used in present study and the larger Sleep and Autism study. Information collected from sleep diary and video recordings were used to calculate sleep problem severity scores.

Table 1

Summary of Participant Characteristics

Characteristics	Values (percentages)
Total number of participants	31
Age range	3-14 years old
Mean Age	6 years and 8 months
Male	25 (81%)
Participants with more than one diagnosis	8 (26%)
Night Wakings	10 (32%)
Co-sleeping	9 (29%)
Sleep Onset Delay (SOD)	7 (23%)
Early Morning Waking	3 (10%)
Parental Presence during SO	8 (26%)
Bedtime Resistance	2 (7%)
Multiple parent-reported sleep problems	10 (32%)

Sleep diary. Sleep diaries were used by parents or caregivers to record daily sleep data. The data included (1) daytime sleep (setting, time asleep, time awake), (2) setting of night time sleep, (3) time put to bed, (4) frequency of curtain calls (i.e. child's bids for parental attention after the child was put to bed), (5) description of the curtain calls, (6) parent's responses to child's curtain calls, (7) estimated time of initial sleep onset, (8) time, frequency and duration of night wakings as well as a description of child's behaviour during night wakings and parents or caregiver responses to the behaviours, and (9) morning wake-up time. Information obtained from the sleep diary was used to determine bedtime at night, wake up time, number of nights spent co-sleeping, parental presence during sleep onset and following night waking. The information was then used to calculate sleep onset latency in minutes (i.e., the difference between bedtime and time till silence), duration

of night wakings in minutes, frequency of night wakings, duration of early morning wakings, and total sleep time (i.e., the difference between time till silence and wake-up time). Sleep diaries were recorded by parents or caregivers for a minimum of two weeks during the assessment phase, and then every night during the baseline, intervention, and follow-up phases of the study. A copy of standard template used for sleep diary is included in Appendix E.

Sleep diaries were collected at least once a week during the assessment phase in order to ensure the diaries were completed accurately, to inform the development of the treatment, and to confirm the eligibility for the study. Sleep diaries were also collected at least once per week during the baseline phase. However, during intervention phase, daily contacts were made with the families to collect sleep diaries in order to inform and to allow for prompt changes to the intervention strategies if necessary. During follow-up, sleep diaries were collected at the end of follow-up period which typically ran for a week.

Video recordings. Night-time infra-red video recordings were used to capture child behaviours after they were put to bed. The video equipment contained adequate storage capacity to record for the duration of the night. Video recordings were collected for a minimum of 30% of baseline, intervention and follow-up phases of the study. In the present study, video recordings were used to supplement data from sleep diaries and used, as part of the FBA process, to analyse antecedent and consequence variables that might be maintaining the sleep problems.

Inter-Observer Agreement (IOA). In order to obtain IOA, information recorded in the sleep diaries were compared with the video recordings. This task was performed by a research assistant who was blind to the sleep diaries. At least 20% of the video recordings were viewed by the research assistant across each phase of the study and information was recorded on a separate sleep diary similar in format to the sleep diary completed by the family. In terms of frequency and occurrence of the behaviour, agreement was recorded if both the parent and the observer observed the occurrence and frequency of the behaviour. Disagreement was recorded if only one party observed the behaviour. In terms of duration of observed behaviours, agreement was recorded if

both parent and observer's recorded duration were within 15 minutes of each other. A percentage of agreement for each behaviour was then calculated using the equation $[\text{Agreements}/(\text{Agreements} + \text{Disagreements})] \times 100$.

The Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000).

The CSHQ was administered during baseline and post-treatment phase. The CSHQ is a 33-item rating scale designed to assess the type and severity of sleep problems in children aged 4-10 years. It contains items related to eight sleep subscales: (1) Bedtime Resistance, (2) Sleep Onset Delay, (3) Sleep Duration, (4) Sleep Anxiety, (5) Night Wakings, (6) Parasomnias, (7) Sleep Disordered Breathing, and (8) Daytime Sleepiness. Parents or caregivers report the frequency of particular sleep behaviours observed over the previous week on a three point scale: *Rarely* (0-1 night per week), *sometimes* (2-4 nights per week), or *usually* (5-7 nights per week). In addition, parents or caregivers are requested to indicate whether these particular behaviours are a problem for the family. A total sleep disturbance score is then calculated together with the eight subscale scores relating to specific sleep disturbances.

Owens and colleagues (2000) reported an internal consistency of 0.68 and 0.78 for the community sample ($n = 469$) and clinical sample ($n = 154$), respectively. Test-retest reliability was acceptable (range 0.62 to 0.79). Receiver operator characteristic (ROC) analysis identified a total score of 41 as the clinical cut-off for sleep problems with a sensitivity of 0.80 and specificity of 0.72. (Owens et al., 2000). Goodlin-Jones and colleagues (2008) showed that the CSHQ discriminated between good and poor sleepers in a sample of 194 preschool children, typically developing and those with ASD, down to 2 years of age (Goodlin-Jones et al., 2008).

For the purpose of the present analysis, CSHQ total raw scores were used as a measure of dimensions for child clinical complexity and used to identify latent factors predicting the responses to treatment. Higher CSHQ scores indicate higher parent-reported sleep problems.

Measures of Sleep Problem Severity. Following McLay, France, Knight, Blampied and Hastie (2019), a measure of Sleep Problem Severity (SPS) was formulated using an adapted version

of the Sleep Behaviour Scale (SBS; Richman, 1985). The SPS scoring system was adapted to incorporate developmental norms regarding bedtimes and total sleep time from the National Sleep Foundation Guidelines (NSF; Hirshkowitz et al., 2015; Ohayon et al., 2017) and include percentage of night spent co-sleeping each night and parental presence for initial sleep onset and parental presence during sleep onset following night waking. The percentage of night spent co-sleeping each night has five categories (0, 1, 2, 3, 4), where 0 was considered to be developmentally appropriate and 4 was considered to contributing to significant sleep problems. Parental presence for initial sleep onset or parental presence during sleep onset following night waking was dichotomy (0, 1), where 0 was considered to be developmentally appropriate and 1 was considered to be contributing to sleep problems. The items on parental presence are added to reflect the strong negative association between total sleep time and parental presence for initial sleep onset as well as the association between parental presence and increased night wakings. Items from the original SBS included average time taken to sleep or average bedtime (whichever is worse), average total time slept at night, average numbers of night waking per week, average number of wakings per night, average time awake per waking, and average weekly hours in parents' bed. Each item has five categories (0, 1, 2, 3, 4), where 0 was considered to be developmentally appropriate and 4 was considered to be having significant sleep problems.

The SPS Scoring system was adapted for three different age groups, to reflect developmental norms: preschool-aged children (aged 3 to 4 years and 11 months old), primary school-aged children (aged 5 to 12 years old) and teens (aged 13 years old and above). Detailed developmentally-appropriate items for SPS score calculation for pre-school, primary school-aged, and adolescents are attached in Appendix F to H. SPS scores were calculated for the final seven days of baseline and treatment, and all seven days of short- and long-term follow-up (where available). Average of the seven daily SPS score (daily score summed over 7 days and then divided by 7) was considered as the SPS score for each week. The clinical direction of change for SPS scores is to reduce if sleep improves after completion of treatment. Data for the SPS score was

predominantly obtained from sleep diaries though on occasion, it was necessary to supplement this with video recordings (e.g., if insufficient data was recorded in the sleep diary).

Measures of children's daytime functioning and behaviour.

The following measures were administered in the larger Sleep and Autism study and the data obtained were examined and analysed in the present study to inform child-specific dimensions of clinical complexity.

Child Behaviour Checklist (1.5-5 years, 6-18 years) (CBCL; Achenbach & Rescorla, 2001). The total CBCL scores are used to provide an assessment of the severity of dysfunction across a broad range of symptom domains and has been used frequently in assessment and intervention studies (Kazdin & Whitley, 2006). The CBCL was used to assess child psychopathology, social competence, and daytime behaviour. The CBCL is a questionnaire completed by parents or caregivers based on their knowledge and observations of their child's daytime behaviour. The CBCL examines psychopathology and daytime behaviour across eight domains (anxious/depressed, somatic complaints, withdrawn/depressed, aggressive behaviour, rule-breaking behaviour, thought problems, social problems, and attention problems) and is used to calculate sub-domain scores (Internalizing, Externalizing, and other syndrome scales). There are two versions of the CBCL: preschool (age 1.5 to 5 years old) and school age (aged 6 to 18 years old). Mean test-retest reliabilities of $r = .85$ and $r = .88$ across eight day period for the preschool and school age forms, respectively (Achenbach & Rescorla, 2001). The CBCL T-score was used as a measure of ASD symptom severity. By definition, T-scores of less than <65 ($<93^{\text{rd}}$ percentile) are in the normal range, between 65 and 70 (93^{rd} - 98^{th} percentile) are in the borderline clinical range, and ≥ 70 ($\geq 98^{\text{th}}$ percentile) are in the clinical range (Bruni et al., 2007; Goldman et al., 2009). CBCL was administered during the baseline and maintenance phase of the larger Sleep and Autism study. In the present study, the CBCL T-score was considered as dimensions of child clinical complexity measures and used to identify latent factors predicting the responses to treatment.

The Gilliam Autism Rating Scale, Second Edition (GARS-2; Gilliam, 2006). The GARS-2 was used to identify ASD and to evaluate the severity of autism symptomology based on the diagnostic criteria of the DSM-IV-TR. The GARS-2 is a norm-referenced 42-item, parent or caregiver report, screening questionnaire. It is appropriate for use with individuals between the ages of 3 and 22 years. The GARS-2 contains three subscales – Communication, Stereotyped Behaviours, and Social Interaction. Each item is rated on a four-point Likert scale (0 = “Never Observed”, 1 = “Seldom Observed”, 2 = “Sometimes Observed”, and 3 = “Frequently Observed”). Total raw scores on the 14-item subscales are converted to standard scores. Based on the total standard scores from the Stereotyped Behaviour and Social Interaction subscale, a norm-referenced total score, the Autism Index (AI; $M=100$, $SD = 15$) is calculated. The Autism Index is used to classify the risk for autism. AI scores ≤ 69 suggest that autism is “unlikely”. AI scores between 70 and 84 suggest that autism is “possible”. AI scores ≥ 85 suggest that autism is “very likely”. Evidence of the reliability and validity of the GARS-2 indicates that internal consistency estimates were .86 for Communication, .84 for Stereotyped Behaviours, .88 for Social Interaction, and .94 for the Autism Index. Corrected test-retest coefficients (1-week interval) based on parent ratings of 37 children with ASD were .70 for Communication, .90 for Stereotyped Behaviours, .88 for Social Interaction, and .88 for the overall Autism Index. Using a cut-off score of 85 for the GARS-2 Autism Index, sensitivity and specificity for the ASD group versus typical controls were 1.00 and .87, respectively (Gilliam, 2006). The GARS-2 classification of autism risk was used as a measure of autism symptom severity. The autism severity level was coded as two when, based on GARS-2 convention, autism was ‘possible’ and the autism severity level was coded as a three when autism was ‘very likely’. The GARS-2 was administered during baseline and maintenance phase of the larger Sleep and Autism study. In the present study, the GARS-2 Autism Index level was considered as dimensions of child clinical complexity measures and used to identify latent factors predicting the responses to treatment.

The Vineland Adaptive Behaviour Scales, Second Edition (VABS-II; Sparrow, Cicchetti, & Balla, 2005). The VABS-II is used to assess the adaptive behaviour of individuals from birth through to adulthood. For the Sleep and Autism study, only the VABS-II receptive and expressive communication sub-domains were administered. Items on the VABS-II are rated on a scale from zero to two with zero representing *behaviour never performed*, one representing *behaviour sometimes or partially performed*, and two representing *behaviour usually or habitually performed*. Domain scores and the composite scores are standardized ($M = 100$, $SD = 15$). Test-retest reliability for the VABS-II has been established; subdomain reliability coefficients are excellent with most values exceeding .85 (Sparrow et al., 2005). The VABS-II was used during the assessment phase in the larger Sleep and Autism study to determine the child's level of receptive and expressive language, to confirm their eligibility for the study, and to tailor the interventions to the child's level of understanding. In the present study, the VABS-II v-scale score was considered as dimensions of child clinical complexity measures and used to identify latent factors predicting the responses to treatment.

Other measures.

In the present study, types of comorbid psychopathology were consolidated into categorical variable of comorbidity. Types of medications were consolidated into categorical variable of medications.

Comorbidity. Comorbidity was calculated based upon the number of *Diagnostic and Statistical Manual of Mental Disorders (5th ed.; DSM-5; American Psychiatric Association [APA], 2013)* diagnoses other than ASD. For example, Global Developmental Delay (GDD), Attention Deficit Hyperactivity Disorder (ADHD), Dissociative Disorders (DD), Tic Disorders, Specific Learning Disorder (SLD), Dyscalculia, and Developmental Coordination Disorder (DCD)/Dyspraxia. Due to the relatively small number of children having additional diagnosis, the variables were collapsed into the following categories (0, 1, 2, 3), where 0 was designated for ASD diagnosis only, 1 for ASD and other sleep comorbidities (i.e., delayed sleep onset latency, co-

sleeping, frequent or prolonged night wakings, early morning wakings), 2 for ASD and non-sleep comorbidities, and 3 for both sleep and non-sleep comorbidities in addition to ASD. In the present study, the category of comorbidity was included as one of the measures used to identify latent variables that would serve as a dimension of child clinical complexity.

Medications. Given that medications and behavioural treatments might synergistically moderate each other (Aman et al., 2009) or behavioural intervention effects and medication effects might be additive in a straightforward manner (Scahill et al., 2012), medications were included in the analysis. Medication information was obtained through review of individual case information and was included as one of the variables in factor analysis. In present sample, medications taken include selective serotonin reuptake inhibitors (e.g., Fluoxetine), selective norepinephrine reuptake inhibitor (e.g., Atomoxetine), hormones (e.g., melatonin), antipsychotics (e.g., Risperidone), central nervous system stimulant (e.g., Concerta) and antihistamines (e.g., Trimeprazine, Phenergan, Vallergan). No children taking anti-epilepsy medications or asthma medications was included in the present study. The use of medication was categorised and scored as follows: 0 = no medication, 1 = melatonin, 2 = use of psychotropics, and 3 = use of both melatonin and psychotropic medications. In the present study, the category of medications was included as one of the measures used to identify latent variables that would serve as a dimension of child clinical complexity.

Latent variables of child clinical complexity.

In the present study, following Kazdin and Whitley (2006), data from baseline measures of children's daytime functioning and behaviour (i.e., CBCL, GARS-2, and VABS II), measures of children's sleep problem (i.e., CSHQ), categorical variable of comorbidity and categorical variable of medications were used to identify latent variables that would serve as dimensions of clinical complexity.

Data Analysis

Existing clinical data from 31/42 participants in the Sleep and Autism study was analysed. Age, gender, comorbidity, prescribed medications, v-Scale scores of the receptive and expressive

communication sub-domains of the VABS II, T-scores of the CBCL, raw scores of CSHQ, severity level scores of GARS-2, and scores of SPS were entered into an Excel spreadsheet. Four data analysis steps were employed in present study. These steps were (1) descriptive and exploratory analysis, including bivariate correlations; (2) treatment outcome analysis; (3) factor analysis relating to child complexity; and (4) multiple regression to examine the relationship of any complexity factors to treatment outcome. All statistical calculations were performed using the Statistical Package for the Social Sciences software, *SPSS Version 25* (IBM Corporation, Released 2017).

Descriptive and exploratory analysis.

The descriptive statistics were calculated for the research sample. This included means, standard deviations, the range and the shape of the frequency distributions, as measured by skewness and kurtosis, and clinical cutoff values for any measures where available. Descriptive statistics are useful for investigating characteristics of data and examining the validity of statistical assumptions underlying inferential multivariate statistical test (Meyers, Gamst, & Guarino, 2006). In descriptive statistics, measures of skewness and kurtosis can be used to analyse the normative distribution of data and to examine minimum, maximum, and mean values (Grant, Ries, & Thompson, 2016).

In the present study, means and standard deviations were calculated for all the variables based on data recorded in sleep diaries. Specific descriptive statistics were chosen to screen data and to assess the normality of data distribution using both quantitative measures of skewness and kurtosis and graphical presentation. Skewness is a measure of the asymmetry in a data distribution and is quantified by the extent to which a data distribution differs from a normal distribution. Kurtosis is a measure of the height of the peak of a probability distribution. A standard normal distribution has kurtosis of 3. Any kurtosis bigger than 3 can be visualized as a thin bell-shaped distribution with a high peak and thin distribution tails. Kurtosis smaller than 3 corresponds to a broad bell-shaped distribution with a low peak and thick distribution tails. Both skewness and kurtosis may serve as indicators of normality (Kallner, 2018). For a normally distributed variable,

the skewness values and kurtosis values will be close to zero (Meyers et al., 2006). A rule of thumb to test whether the departure from normality is not too extreme is to divide the skewness and kurtosis values by the corresponding standard error and the results should not be greater than ± 1.96 (Rose, Spinks, & Canhoto, 2015).

Pearsons Correlations. Pearson correlations were computed to determine the relationship between pre-treatment scores for all complexity and comorbidity measures used in the study. Pearson correlation is commonly used to determine how closely two variables are related. Correlation coefficients (r) can vary from -1 to 1. A positive r denotes that an increase in the value of one variable would lead to an increase in the other variable. Correlation coefficients as high as 1 or -1 are very rare (Emerson, 2015). Any r value between .70 and .90 are considered high, those between .50 and .70 are considered moderate and those between .30 and .50 are considered low. Any r value between .00 and .30 are negligible (Mukaka, 2012).

Treatment Outcomes Analysis.

Statistical analysis was undertaken to determine whether treatment altered the child's sleep to a clinically significant degree. The first step in the analysis was to provide descriptive statistics on the independent variables obtained pre- and post-treatment. Examples of these descriptive information are means, standard deviations, range, and distributions. Results from paired sample t tests was then used to determine whether the differences pre- and post-treatment were statistically significant. A commonly used effect size, Cohen's d , was then calculated to quantify the improvements observed pre- and post-intervention. Effect size d is based on the difference between observations, divided by the average standard deviation of these observations (for within-sample data). An effect size of 0.2 is considered small, those between 0.2 and 0.5 are considered as medium, those between 0.5 and 0.8 as medium to large, and those > 0.8 are considered large (Cohen, 1988). Effect size estimates provide a more generally interpretable description of the size of observed effects that is independent of the possibly misleading influences of sample size (Fritz, Morris, & Richler, 2012).

Changes over time in SPS scores for each case were plotted as a modified Brinley plot (Blampied, 2017). Modified Brinley plots were used to identify systematic effects of treatments. On modified Brinley plots, all points directly on or clustered about the 45° diagonal line signal no treatment effect. Systematic movements of points above or below the diagonal line signal either improvement or deterioration for some or all participants. Modified Brinley plots complement other forms of analysis because of the ability to show direction and extent of change for each participant relative to both that individual's pre-treatment state and that of other participants, thus permitting assessment of the consistency with which outcomes are replicated (Blampied, 2017) case by case.

Factor Analysis.

Child complexity is a latent variable and is not directly observable. Exploratory factor analysis was, therefore, used to detect any unobservable (latent) variables representing the complexity construct (Zygmunt & Smith, 2014) and also used to summarize and combine the seven measures (number of medications, comorbidity, VABS receptive, VABS expressive, CBCL, GARS severity level, and CSHQ) into these underlying latent variables. In order to determine the factorability of an intercorrelation matrix, two tests were conducted; the Kaiser-Meyer-Olkin measure of sampling adequacy and Bartlett's test of sphericity. The Kaiser-Meyer-Olkin measure of sampling adequacy indicates the proportion of variance in the variables that might be caused by underlying factors ("IBM Knowledge Center," n.d.). Acceptable values for the Kaiser-Meyer-Olkin measure of sampling adequacy range from 0.5 to 1 (Hutcheson & Sofroniou, 1999). Any value less than 0.5 indicates that the results of the factor analysis probably won't be very useful ("IBM Knowledge Centre," n.d.). Bartlett's test of sphericity indicates whether the correlation matrix is an identity matrix, in which all the diagonal elements are 1 and all off diagonal elements are 0 ("UCLA: Statistical Consulting Group," n.d.). A significance level of less than 0.05 indicates that a factor analysis may be useful with the data ("IBM Knowledge Centre," n.d.). If both the value of Kaiser-Meyer-Olkin measure of sampling adequacy is less than 0.5 and the significance level of the Bartlett's test of sphericity is much more than 0.5, then factor analysis would not be useful.

The factor analysis step used in the study involved a Principal Component Analysis extraction method using Varimax with Kaiser Normalization rotation method. Principal Component Analysis is a way of identifying trends and patterns in data with the goal to highlight similarities and differences (Smith, 2002). The criterion for terminating the number of factors being extracted in the analyses was an eigenvalue of 1.0 (Grant, Ries, & Thompson, 2016). The goal of Principal Component Analysis is to explain as much of the variance in the matrix of raw scores using as few components as possible. To allow for theoretical interpretation, varimax rotation is used to clarify the unrotated factor loading matrix (Zygmunt & Smith, 2014). In Varimax rotation, the variance accounted for by each of the factors is maximized and the total amount of variance accounted for is redistributed over the three extracted factors (“UCLA: Statistical Consulting Group,” n.d.). Based on the results of factor analysis, the scores of the latent variables, commonly called factor scores, can be predicted. In present study, regression predictors (Thomson, 1934; Thurstone, 1935, as cited in Devlieger, Mayer, & Rosseel, 2015) are used to compute the factor scores to be used in the subsequent linear regression. Note that if child complexity is a single, real construct, the expectation is that there would be a single factor extracted by the factor analysis.

Multiple Regression.

As the final step of data analysis, child’s age, gender, and the one of more factor scores obtained from the factor analysis were used as predictors of treatment responses. A hierarchical multiple regression is performed in order to control for the effects of covariates and to test the effects of certain predictors independent of the influence of others. In the hierarchical multiple regression, sociodemographic variables (e.g., age and gender) were entered into the first block, and the child complexity factor scores are entered jointly into the second block of the linear regression.

Chapter 4

Results

This chapter presents the descriptive statistics of pre-treatment scores on the VABS receptive and expressive subscale, CBCL, CSHQ and SPS and post-treatment SPS scores. A Pearson correlation matrix is presented to determine the associations among age, gender, and all pre-treatment and post-treatment scores. Modified Brinley plots are presented for pre- and post-treatment SPS scores. Model summaries from multiple regression analyses are also presented.

Information on age, gender, medication, comorbidity, pre- and post-treatment SPS scores were available for the 31 children. For 24 out of 31 children, parents or caregivers completed the GARS, VABS receptive and expressive subscale, CBCL, and CSHQ. Two children were siblings and had the same parents.

Quality of Data

The parents or caregivers of 3 children did not complete VABS Receptive and Expressive scores. The CBCL was not administered to 6 out of 31 children. The GARS was not administered to 7 out of 31 children. Pre-treatment CSHQ scores were not available for 5 out of 31 children. Out of these 5 children, the CSHQ could not be administered to one of the children because the child's age was outside of the validated range for the CSHQ. Lastly, three children were administered the CSHQ abbreviated version.

Child VABS Receptive and Expressive, CSHQ, CBCL Measures

Table 2

Descriptive Statistics for Children Pre-treatment Measures

Measures	<i>N</i>	<i>M</i>	<i>SD</i>	<i>Range</i>	<i>Min</i>	<i>Max</i>	<i>Skewness</i>		<i>Kurtosis</i>		<i>Skewness</i>	
							<i>Statistics</i>	<i>Std. Error</i>	<i>Statistics</i>	<i>Std. Error</i>	<i>Statistics</i> <i>Std. Error</i>	<i>Statistics</i> <i>Std. Error</i>
VABS Receptive	28	8.61	2.17	8.0	5.0	13.0	0.31	0.44	-0.40	0.86	0.70	-0.47
VABS Expressive	28	9.29	2.68	11.0	5.0	16.0	0.58	0.44	0.64	0.86	1.32	0.74
CBCL total	25	68.48	11.0	49.0	43.0	92.0	-0.41	0.46	0.73	0.90	-0.89	0.81
CSHQ	26	51.54	8.50	33.0	36.0	69.0	-0.06	0.46	-0.35	0.89	-0.13	-0.39

Note. *N* = Participants, *M* = Mean, *SD* = Standard Deviation, *Min* = Minimum, *Max* = Maximum

The Relationship between the VABS Receptive, VABS Expressive, CBCL, and CSHQ

Table 3

Pearson Product Moment Correlation of pre-intervention psychometric scores

Variable	1	2	3	4	5	6	7
1. Medication	1	0.29	-0.382*	-0.21	0.07	0.11	-0.03
2. Comorbidity		1	-0.18	0.23	0.34	-0.35	0.12
3. VABS Receptive			1	0.563**	0.06	-0.02	-0.17
4. VABS Expressive				1	0.35	-0.28	-0.05
5. CBCL Total					1	-0.18	0.26
6. GARS Level						1	0.09
7. CSHQ							1

Note. Correlations printed in bold are significant; * $p < .05$., ** $p < .01$.

Child SPS Measures

Table 4

Descriptive Statistics for Children SPS Pre- and Post-treatment Scores

Measures	<i>N</i>	<i>M</i>	<i>SD</i>	<i>Range</i>	<i>Min</i>	<i>Max</i>	<i>Skewness</i>		<i>Kurtosis</i>		<i>Skewness</i>		<i>Kurtosis</i>	
							<i>Statistics</i>	<i>Std. Error</i>	<i>Statistics</i>	<i>Std. Error</i>	<i>Statistics</i>	<i>Std. Error</i>	<i>Statistics</i>	<i>Std. Error</i>
Baseline	31	5.61	2.69	11.0	1.0	12.0	0.42	0.42	-0.09	0.82	1.00	1.00	-0.11	-0.11
Post-treatment	31	2.26	1.61	6.00	0.00	6.00	0.57	0.42	-0.26	0.82	1.36	1.36	-0.32	-0.32
Short-term FU	23	2.17	1.90	7.00	0.00	7.00	0.73	0.48	0.11	0.94	1.52	1.52	0.12	0.12
Long-term FU	22	1.77	1.69	6.00	0.00	6.00	0.98	0.49	0.58	0.95	2.00	2.00	0.61	0.61

Note. *N* = Participants, *M* = Mean, *SD* = Standard Deviation, *Min* = Minimum, *Max* = Maximum, FU = Follow-up

Descriptive and Exploratory Analysis

Table 2 displays the means, standard deviations, range, minimum and maximum values, skewness and kurtosis statistics for VABS receptive and expressive language, CBCL total behaviour, and CSHQ.

Prior to treatment, the average *v*-Scale score on the VABS Receptive subdomain (8.61) and VABS Expressive subdomain (9.29) were considered low (Sparrow, Cicchetti, Saulnier, 2016). Average CBCL total behaviour problem score (68.48) was in the borderline clinical range. The average score on the CSHQ (51.54) was within the clinical range. The results of dividing skewness and kurtosis value by its corresponding standard error were also shown on Table 2. The results were all well within ± 1.96 limits, suggesting that the departure from normality is not too extreme.

Pearson product-moment correlation coefficients were computed to determine the association between pre-treatment scores used in the study. These correlations are presented in Table 3. Statistically significant correlations are observed between three variables. These correlations were large enough to reject the null hypothesis that the true value of r in the population = 0. Medication had a low negative correlation with VABS Receptive scores. As expected, VABS Receptive had a moderate positive correlation with VABS Expressive scores.

A weak but significant negative correlation was found between medication and VABS Receptive scores indicating increased use of melatonin and/or psychotropic medications was correlated with lower VABS Receptive score. This correlation was in the direction expected because while the use of psychotropic medications might not be effective for the core features of ASD (social and communication impairment) (Madden et al., 2017), the use of melatonin alone seemed to improve communication (Wright et al., 2011). Hence, the positive impact of melatonin on communication might be diluted by the lack of impact of psychotropics on communication since the present study combined both psychotropics and melatonin into the categorical variable of medication.

A significant strong positive correlation was found between VABS Receptive and VABS Expressive scores indicating difficulty in receptive communication domain was correlated with difficulty in expressive communication domain. This correlation was in the direction expected because individual challenges in using word and sentences to gather and provide information would be associated with challenges to listen, to pay attention and to understand.

No significant correlations were found between medication and comorbidity, VABS Expressive, CBCL, GARS level, or CSHQ. No significant correlations were found between comorbidity and VABS Receptive, VABS Expressive, CBCL, GARS or CSHQ. No significant correlations were found between VABS Receptive and CBCL, GARS or CSHQ. No significant correlations were found between VABS Expressive and CBCL, GARS, or CSHQ. No significant correlations were found between CBCL and GARS or CSHQ and lastly, no significant correlations were found between GARS and CSHQ.

Treatment Outcomes Analysis

Table 4 displays the means, standard deviations, range, minimum and maximum values, skewness, and kurtosis on the SPS scores at baseline, post-treatment, short-term follow-up and long-term follow-up. SPS scores obtained at baseline, post-treatment and short-term follow-up are normally distributed, as shown by the results of dividing skewness and kurtosis value by their corresponding standard error. Almost all results were well within ± 1.96 limits, suggesting that the departure from normality is not too extreme; however, the results for long-term follow-up SPS scores exceeded the 1.96 limits and seemed to suggest some departure from normality.

Results from paired samples *t* tests indicated that SPS scores were statistically significantly lower post-treatment ($M = 2.26$, $SD = 1.61$) than pre-treatment ($M = 5.61$, $SD = 2.69$), $t(30) = 6.64$, $p < .001$, $d = 1.51$. The effect size was considered large and reliably different from zero (95% CI [2.32, 4.39]). This was also the case at short-term follow-up ($M = 2.17$, $SD = 1.90$) ($M = 5.43$, $SD = 3.01$), $t(22) = 5.08$, $p < .001$, $d = 1.30$ (95% CI [1.93, 4.59]) and at long-term follow-up ($M =$

1.77, $SD = 1.69$) than pre-treatment ($M = 5.68$, $SD = 3.08$), $t(21) = 5.69$, $p < .001$, $d = 1.57$ (95% CI [2.48, 5.34]). At each time point the effect size was large and reliably different from zero.

Modified Brinley Plots Interpretation

As seen in Figure 1, modified Brinley plots show individual changes pre- and post-intervention simultaneously on one graph enabling the identification of systematic effects of an intervention. X-axis represents pre-intervention scores and Y-axis represents post-intervention scores. Each point on the graph represents an individual's data with the X coordinate being the pre-intervention score and Y coordinate being the post-intervention score. A vertical line represents the clinical cut-off for pre-intervention scores and a horizontal line represents the clinical cut-off for post-intervention scores. Together with the 45° diagonal line (i.e., line of no change), the horizontal and vertical clinical cut-off lines partition the graph into five zones. Starting from the lower left-hand corner and moving anti-clockwise, these zones are “Pre: Non-clinical, Post: Non-clinical”, “Pre: Clinical, Post: Non-clinical”, “Pre: Clinical, Post: Clinical but improved”, “Pre: Clinical, Post: Clinical but worsened”, and “Pre: Non-clinical, Post: Clinical”. For those individuals that fall within the first zone “Pre: Non-clinical, Post: Non-clinical”, the interpretation is that the individual's pre- and post-intervention scores both fall below the clinical cut-off line. For those individuals that fall within the second zone “Pre: Clinical, Post: Non-clinical”, the interpretation is that improvement has been achieved since the score, above the clinical cut-off line before treatment, has dropped below the clinical cut-off after treatment. Finally, for those individuals that fall within the last zone “Pre: Non-clinical, Post: Clinical”, the interpretation is that deterioration has occurred since the score, below the clinical cut-off before treatment, has surpassed the clinical cut-off post treatment.

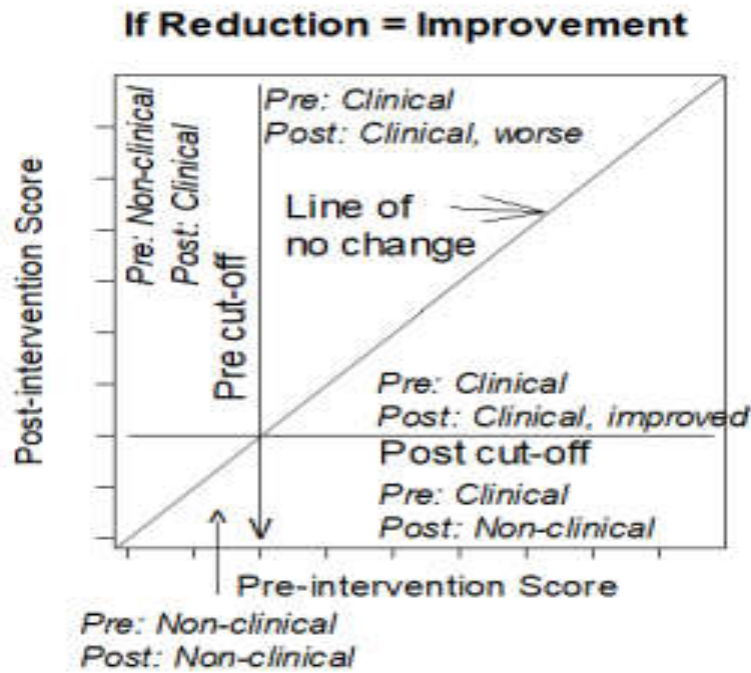


Figure 1. Conventions for interpreting modified Brinley plots where reduction in score represents clinical improvement (Blampied, 2017).

Modified Brinley Plots – SPS scores

The SPS scores are presented below on modified Brinley plots showing individual change from pre- to post-intervention, short-term and long-term follow-up. Given that SPS scores are formulated so that a comparable measure is available to show change, the determination of any clinical cut-off point is beyond the scope of this thesis. Nevertheless, Figure 2 and 3 clearly shows that the baseline SPS scores were much higher than post-treatment, short-term, and long-term follow-up SPS scores. As seen in Figure 2 (A), twenty seven out of 31 children show improvement. The remaining 4 children either did not show any improvement since their data points lie on or near the 45-degree diagonal lines of no change (i.e. no or minor systematic differences between pre- and post-intervention/STFU SPS scores). Similarly, as seen in Figure 2 (B), nineteen out of 23 children show improvement. The remaining 4 children either did not show any improvement since their data points lie on or near the 45-degree diagonal lines of no change.

Finally, similar trends are observed in Figure 3. Nineteen out of 22 children show improvement. The remaining 3 children did not show any improvement since their data points lie near the line of no change.

Factor Analysis

A factor analysis of the child complexity and comorbidity data was performed using the Principal Component Analysis method of factor extraction. Principal Component Analysis arranges the variables into separate factors based on the strength of correlations. To determine whether correlations support factor analysis, the Kaiser-Meyer-Olkin measure of sampling adequacy is calculated. The Kaiser-Meyer-Olkin measure of sampling adequacy indicated that the strength of the relationships among variables was acceptable ($KMO = 0.50$). Bartlett's test of sphericity indicated that the correlations within the correlation matrix was not significant ($\chi^2 (21) = 29.7, ns$).

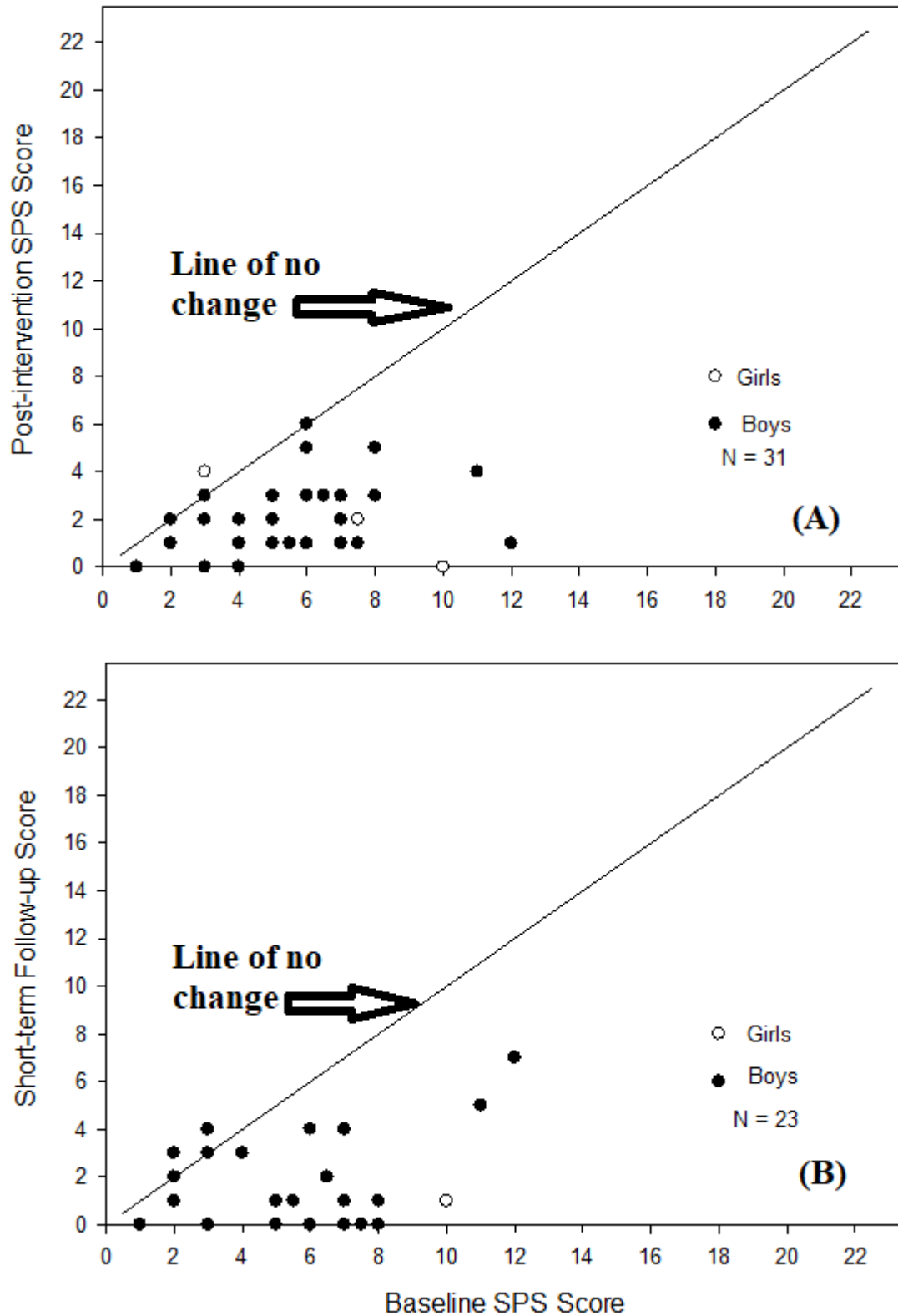


Figure 2. Modified Brinley plot showing change on SPS scores from pre- to post-intervention (A) and short-term follow-up (B).

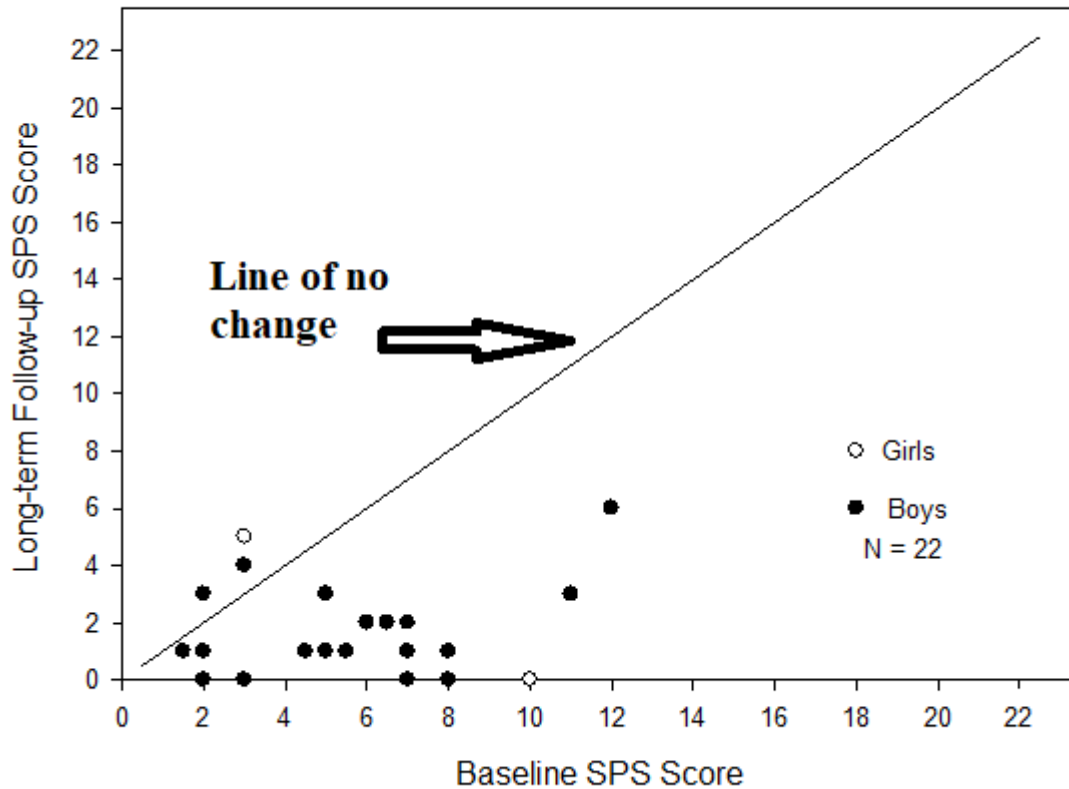


Figure 3. Modified Brinley plot showing change on SPS scores from pre-intervention to long-term follow-up.

However, since results from one of the two test was adequate, it was acceptable to proceed with the analysis.

Three factors with eigenvalues greater than one were extracted. A Varimax rotation with Kaiser Normalization was performed. The obtained rotated component matrix is displayed in Table 5. After Varimax rotation, the first factor was robust, with an eigenvalue of 2.02 and accounted for 28.8% of the variance in the data. The second factor had an eigenvalue of 1.59 and accounted for an additional 22.7% of the variance in the data. The third factor had an eigenvalue of 1.40 and accounted for an additional 20.0% of the variance in the data. Cumulatively, the three factors accounted for a total of 71.5% of the variance in the data. Factor analysis of the child complexity and comorbidity items used in the present study revealed three factors were sufficient to explain the underlying latent variables.

Table 5

Rotated Component Matrix for child complexity factors

Measures	Factor I	Factor II	Factor III
Medication	-0.71	0.29	-0.04
Comorbidity	-0.31	0.72	0.30
VABS Receptive	0.86	-0.04	-0.23
VABS Expressive	0.79	0.40	0.13
GARS Level	-0.10	-0.79	0.12
CSHQ	-0.17	-0.18	0.88
CBCL	0.16	0.40	0.67

Bold-face values indicate the factor that the variables was adopted onto.

The factors were named as follows: I. *Medication-Communication* (Medication, VABS Receptive and VABS Expressive), II. *Psychopathology Severity* (Comorbidity and GARS level), III. *Behaviour and Sleep Problems* (CSHQ and CBCL). Overall, the factor analysis revealed that all items loaded onto these three factors.

Multiple Regression

Table 6 presents the results from hierarchical multiple regression analyses using the treatment outcome scores as predicted variables. Age and gender were entered at Step 1. At Step 2, the role of Factor Score 1, 2, and 3 were examined controlling for age and gender. Factor scores were produced using regression approach and were assigned to each child for each of the 3 complexity factors. These factor scores were treated as variables in the hierarchical multiple regression. The hierarchical multiple regression revealed that, at Step 1, Age and Gender contributed significantly to the regression model, $F(1,20) = 3.64, p < .05$) and accounted for 29% of the variation in SPS score changes. Introducing the Factor Score variables explained an additional 21% of variation in SPS score changes and this change in R^2 was significant, $F(1,20) = 3.00, p < .05$). When all five independent variables were included in step 2 of the regression model, only

Factor Score 3 was the significant predictor of changes in SPS scores. Together the five independent variables accounted for 50.0% of the variance in SPS score changes.

Table 6

Summary of Hierarchical Regression Analysis for Variables predicting changes in SPS score

Variable	β	t	R	R^2	ΔR^2	Increment (%) ^a
Step 1			0.54	0.29	0.29	29
Age	0.32	1.53				
Gender	0.35	1.67				
Step 2			0.71	0.50	0.21	21
Age	0.07	0.30				
Gender	0.35	1.71				
Factor Score 1	-0.11	-0.57				
Factor Score 2	0.23	1.10				
Factor Score 3	0.46	2.26*				

Note. $N = 21$; $*p < .05$

^a Represents proportion of variance accounted for by variables at their entry point into regression equation. $R^2/\Delta R^2$ values multiplied by 100.

An additional analysis was conducted on both CSHQ and CBCL scores – components of Factor Score 3. Using the median of CBCL T-scores, the group was split into two – a Low CBCL group and a High CBCL group. Means and standard deviations of the SPS change scores between the two groups were used to calculate the Cohen's d effect size. A medium to large Cohen's d effect size was obtained ($d = 0.62$) when evaluating the differences in mean SPS change scores between the low CBCL and high CBCL group. Similarly, using the median of CSHQ raw scores, the group was split into two – a Low CSHQ group and a High CSHQ group. Means and standard deviations of the SPS change scores between the two groups were also used to calculate the Cohen's d effect size. A small to medium Cohen's d effect size was obtained ($d = 0.38$) when evaluating the differences in mean SPS change scores between the low CSHQ and high CSHQ group. Also,

looking at the standard deviations of the two groups based on either CBCL median split or CSHQ median split, higher CBCL T-scores or CSHQ raw scores are associated with reduced variation in changes in SPS scores. In other words, a comparison of the groups on the change of SPS scores indicated that children with more behaviour or sleep problems showed less change in the SPS scores. Nevertheless, children with more sleep problems still responded reasonably well as measured by the change in SPS scores and the effect size and changes for these children were medium to large. For children with more behaviour problems, there were responses to treatment, however, the effect size and changes for these children were small to medium.

Chapter 5

Discussion

The present study had three research aims: (1) to investigate whether a multi-component, parent-implemented, FBA informed treatment would result in a reduction in sleep problems; (2) to investigate the existence of latent complexity variables that may affect children's responses to treatment; and (3) to investigate whether the latent complexity variables and child characteristics (i.e., gender, age, autism core symptoms severity, co-morbid psychopathology, sleep problems severity, behaviour severity, communications and medications) serve as predictors of treatment response.

In the present study, the implementation of a multi-component, parent-implemented, FBA informed treatment resulted in a reduction in sleep problem severity, as calculated using the SPS scoring system. In addition, out of the seven dimensions of child complexity that were analysed (Medication use, presence of comorbidities, VABS Receptive and Expressive language scores, GARS symptom severity level, CSHQ and CBCL scores), three latent complexity variables were identified. These latent complexity variables were named *Medication-Communication* (as measured by the types of medication and the VABS communication sub-domain), *Psychopathology Severity* (as measured by comorbidity categories and the GARS severity level), and *Behaviour and Sleep Problems* (as measured by CSHQ and CBCL). Only one of the latent complexity variables underlying sleep and behaviour problems served as predictors of treatment response, as measured by changes in SPS score. These findings will be discussed in detail in the following sections. An overview of the strengths and limitations, clinical implications and recommendations for future research will also be provided.

Reductions in sleep problems following multi-component, parent-implemented, FBA-informed treatment

Following treatment, a significant reduction in SPS scores was observed for 26 out of the 31 participants. This change was also reflected in a reduction in CSHQ scores for 19 out of the 22 participants who also had post-treatment CSHQ scores. Prior to the intervention, 24 children (77%) met the CSHQ criteria for the presence of a sleep disorder. When assessed with CSHQ following intervention, 10 of the 24 children no longer met this criterion, and another seven of the children had a reduction in their CSHQ score. In other words, improvements were observed in 71% of those children who originally met the CSHQ criteria of sleep disorder. These findings suggest that a multi-component, FBA-informed, parent-implemented intervention is effective in reducing sleep problems in children with ASD.

The results of this study add to the existing literature by providing further support for the use of individualized FBA informed interventions to treat a range of common sleep problems in children with ASD (Jin et al., 2013; McLay et al., 2017; 2019). Whilst there is a large body of evidence to support the use of behaviourally-based treatments for sleep problems in children with ASD (Cuomo et al., 2017; Mindell et al., 2006; Turner & Johnson, 2012; Vriend et al., 2011), this is one of few studies to have used FBA to inform the formulation of treatment plans (Jin et al., 2013; McLay et al., 2017; 2019). Treatment plans formulated based on the outcomes of FBA are likely to be more effective than treatments that are not informed by the behavioural function, because the treatment plans will address the individual-specific variables that precipitate and maintain the sleep problem (McLay et al., 2017).

Another important finding of this study is that treatment gains were maintained at both short- and long-term follow-up for the majority of participants. The stability of treatment effects was assessed both 4-6 weeks (i.e. short-term) and 10-12 weeks (i.e. long-term) post treatment and it was found that treatment outcomes were further enhanced or maintained at short-term follow-up for 18 out of 23 children and at long-term follow-up for 18 out of 22 children who provided data for

SPS score calculation. This is one of few studies that has examined both short- and long-term maintenance of treatment effects. The majority of existing research on parent-implemented behavioural sleep interventions includes a much shorter follow-up period of 2 to 8 weeks (e.g., Austin, Gordon, & O'Connell, 2013; Christodulu & Durand, 2004; Fawkes, Malow, Weiss, Reynolds, & Loh, 2015; Knight & Johnson, 2014; Loring, Johnston, Shui, & Malow, 2018; Moss, Gordon, & O'Connell, 2014; Scantlebury, et al., 2018) or no follow-up information (e.g., Cortesi, Giannotti, Sebastiani, Panunzi, & Valente, 2012; Delemere & Dounavi, 2018; Durand & Christodulu, 2004; Johnson et al., 2013; Malow et al., 2014; Reed et al., 2009; Schoen, Man, & Spiro, 2017). Long term maintenance of treatment effects is crucial for identification of effective interventions (Kirkpatrick, Louw, & Leader, 2019).

Latent complexity variables that may affect children's responses to treatment

In order to detect any unobservable (latent) variables that may underpin the construct of complexity, an exploratory factor analysis of the presence of such latent variables underlying comorbidities, medication use, sleep problem severity (CSHQ), and measures of children's daytime functioning and behaviour (i.e., CBCL scores, GARS symptom severity level, VABS Receptive and Expressive language scores) was conducted. This analysis yielded three factors relating to three dimensions of clinical complexity: a medication-communication domain, psychopathology severity domain, and behaviour and sleep problem severity domain. The medication-communication complexity domain was negatively correlated with medication and positively correlated with communication suggesting that the present sample was less complex in terms of medication, and more complex in terms of communication. Closer examination of the samples in the present study revealed that 20 out of 31 children in the present study did not take medications. Hence, the majority of the sample were at the least complex end of the medication-communication complexity continuum. In contrast, 26 out of 28 children had moderately low or low levels of expressive and receptive communication skills and thus, were on the complex end of the medication-communication complexity continuum. The psychopathology severity domain factor was negatively

correlated with GARS level and positively correlated with comorbidity suggesting that the present sample was less complex in terms of comorbidity, and more complex in terms of GARS severity level. An examination of the data revealed that only 25% of the children had comorbid psychopathology. Thus, in terms of comorbidity, the sample was on the least complex end of the psychopathology severity continuum. In contrast, 75% of the children had a GARS severity level of 2 or 3. Therefore, in terms of GARS severity level, the sample was on the complex end of the psychopathology severity continuum. Behaviour and sleep problems severity was positively correlated with both daytime behaviours, as measured on the CBCL, and sleep problems, as measured on the CSHQ suggesting that the present sample was complex both in terms of behaviour and sleep problems. Closer observations of the sample revealed that 75% of the children had CSHQ sleep problem severity scores that were in the clinical range and 61% of the children had a borderline or clinical CBCL scores. Thus, all the children were on the complex end of the behavioural and sleep problems complexity continuum. These results simply showed limited variability across all the measures taken from present sample.

Predictors of treatment response

In order to test the independent effects of a single predictor variable on treatment outcomes, independent of the influence of other predictors, a regression analysis was performed. Regression models that adjusted for age and sex suggested that change in severity of sleep problems was associated with severity of daytime behaviour and CSHQ scores. A clinical complexity dimension which was comprised of behaviour problems (as measured by CBCL) and sleep problems (as measured by CSHQ) emerged as a significant predictor of change in SPS scores pre- and post-treatment. The results of the present study are consistent with the findings reported by Remington et al. (2007) in one of the most rigorously controlled 2-year longitudinal field study confirming the effectiveness of intensive behavioural intervention for children with ASD. Children with ASD who respond better have a greater severity of behaviour problems at baseline (Remington et al., 2007). Furthermore, the results of this study are also consistent with previous findings which suggest that

higher rates of challenging daytime behaviour is associated with higher rates of sleep problems (Allik et al., 2006; Cohen et al., 2014; Goldman et al., 2009; 2011; Malow et al., 2006; Mayes & Calhoun, 2009; Mazurek et al., 2019; Schreck et al., 2004; Tudor et al., 2012; Verhoeff et al., 2018).

Further analysis on the latent variables underlying CBCL and CSHQ scores was conducted by performing a median split to identify both the children who experienced a higher versus lower CBCL and CSHQ scores. A comparison of the groups on the change of SPS scores indicated that children with more behaviour or sleep problems showed fewer changes in SPS scores and changes in SPS scores were more prevalent for children with more sleep problems than for children with more behaviour problems.

The remaining complexity dimensions of age, gender, medication-communication and severity of psychopathology were not significant predictors of changes in SPS scores. The results from the present study showed that age, gender, and latent psychopathology complexity factors comprised of both comorbidity and GARS level were weakly but positively linked with overall improvement however, this relationship was not significant. The latent medication-communication complexity factors comprised of medication and VABS communication was also weakly and negatively linked with overall improvement and the relationship was not significant.

There is some evidence within general behaviour analytic research which suggests that children under 4 years of age respond more positively to behavioural interventions than older children (Flanagan et al., 2012; Perry et al., 2011). Hence, given that the present study had a relatively small sample size ($n = 31$) and 25 children (81%) were older than 4 years old, clear association between age at baseline and changes in sleep severity might not be able to be detected.

The finding that gender was not a significant predictor of changes in SPS scores is unsurprising and is consistent with previous findings which suggest that, with the exception of a single study (Tiura et al., 2017) gender is not predictive of treatment outcome (Anderson et al., 2007; Baker-Ericzen, Stahmer, & Burns, 2007; Flanagan et al., 2012; Kasari et al., 2012; Virues-

Ortega, Rodriguez, & Yu, 2013). The results of the present study are likely to be affected by the small sample of female participants ($n = 6$) since the limited sample of female participants may not be adequate in terms of statistical power to detect any association, a limitation cited also by Tiura et al. (2017). Nevertheless, while few studies have investigated the relationship between gender and treatment outcomes, this is an important area of research as it has implications for the individualization of treatment in children with ASD (Halladay et al., 2015).

The number of medications being taken by study participants was not found to be a significant predictor of changes in SPS scores. This is somewhat surprising given that previous studies reported the positive effects of melatonin in combination with cognitive behavioural therapy in addressing sleep problems in children with ASD (Cortesi et al., 2012; Howes et al., 2018). Nevertheless, past studies also reported various side effects of prescribed medications in children with ASD (Haftkamp et al., 2012; Marcus et al., 2009; McCracken et al., 2002; Posey et al., 2007; Simonoff et al., 2013; Wichniak et al., 2017). Antidepressants with activating properties (e.g., fluoxetine), psychostimulant medications (e.g., atomoxetine) and antipsychotics (e.g., risperidone) may all have a negative effect on sleep (i.e., sleep disruptions, insomnia, early morning waking, sedation or drowsiness). The findings should be interpreted with caution because of the following issues. First, only four children were taking melatonin together with psychotropics. Melatonin or psychotropics only were taken by four and three other children, respectively. The small sample of children taking medications might not allow for adequate statistical power to detect any association. Second, a categorical variable was used to measure the number of medications and not the effect of the medications. The lowest possible category denoted no medication use and highest possible category denoted use of both psychotropics and melatonin. Hence, the potential synergetic or antagonistic effects of antidepressants, stimulant, and antipsychotic medications were all clustered into a single category. This may have prevented the detection of an association between effects of medications and treatment responses.

In the present study, receptive and expressive communication level was not a significant predictor of change in SPS scores. This is somewhat surprising given that previous research has found that IQ combined with language ability is one of the strongest predictors of response to behavioural intervention, i.e., higher adaptive behaviour scores are associated with greater improvement following behavioural intervention (Eaves & Ho, 2004; Eikeseth et al., 2007; Flanagan et al., 2012; 2015; Kim et al., 2016; Magiati, Charman, & Howlin, 2007; Magiati et al., 2011; Perry et al., 2011; Remington et al., 2007; Sallows & Graupner, 2005). Also, past studies had reported both negative effects of sleep problems on adaptive behaviour and negative impact of low adaptive function on sleep problems (Cohen et al., 2017; Krakowiak et al., 2008; Taylor et al., 2012). The current finding should be interpreted with caution as the majority of the children in the sample had a moderately low or low level of expressive and receptive communication. Only two children had an adequate expressive communication level and a further two children had an adequate receptive communication level. As a result, the sample might lack heterogeneity in terms of the range of expressive and receptive communication to allow for detection of any association between communication level and treatment responses. Furthermore, it is important to note that the present study only measured one out of five domains of adaptive behaviour. The VABS scores of the other four domains of adaptive behaviour (i.e., daily living skills, socialization, motor skills, and maladaptive behaviour) were not obtained because the VABS was primarily used to inform the development of developmentally appropriate materials (e.g., social stories) and to better understand the adaptive functioning level of each child. Use of the VABS measure was limited in order to minimise parental burden and was not originally intended to be used to assess any pre- and post-treatment change in adaptive functioning. In retrospect, further domains of the VABS could be used to get an additional measure of adaptive behaviour and social skills.

In the present study, autism severity was not a significant predictor of change in SPS scores. The severity of autism symptoms has rarely been examined as a potential predictor of treatment outcome (Matson & Smith, 2008), and not at all within the sleep context. Furthermore, a variety of

measures of autism severity have been used in previous studies that have included children with ASD, making comparison difficult (Howlin et al., 2009). Overall, the findings of previous research are mixed. The majority of past studies suggest that lower baseline autism scores are predictive of better treatment outcomes (Ben-Itzhak & Zachor, 2007; Eaves & Ho, 2004; Perry et al., 2011; Sallows & Graupner, 2005; Vivanti et al., 2013), however there are noted exceptions to this. For example, Mello et al. (2018) reported that children with more severe autism symptoms, as measured by CARS, did not improve as much following behavioural intervention. The authors suggested that children with more severe symptoms might require a more intensive intervention and/or intervention did not target the salient symptoms of ASD. The results from present study need to be interpreted with caution for several reasons. First, the present study used the GARS-2 as the only measures of autism severity. In comparison to the ADOS-G and ADI-R, which are commonly used in research, there is some evidence to suggest that the GARS/GARS-2 may underestimate the likelihood of ASD and that convergence with gold-standard research diagnostic measures is quite poor. Thus, caution should be applied when utilizing GARS/GARS-2 for both research and clinical purposes (Mazefsky & Oswald, 2006; South et al., 2002; Volker et al., 2016). Second, the present study uses the categorical variable of GARS-2 Severity level as a measure of autism severity. As a consequence, all of the children in the sample had either a GARS severity level 2 or 3. By using categorical data and not continuous variable, some important information may have been lost and the statistical power may have been reduced. In addition, considerable variability captured in the continuous data may have been unnecessarily subsumed within each category (Altman & Royston, 2006). As such, the statistical power to detect any relationship between autism severity level and changes in SPS scores was reduced. A better alternative to using the GARS severity level could be to use a continuous variable and established autism measures, such as ADOS-G and ADI-R, to measure autism severity level (Hill et al., 2001; Klinger & Renner, 2000; Tanguay, 2000; Volker et al., 2016). The continuous variable would allow for greater variability to be captured and offer the ability to capture a weak association.

Finally, comorbidity was not a significant predictor of changes in SPS scores.

Unfortunately, very few past studies have investigated the direct effect of comorbidity on treatment responses. Kazdin and Whitley (2006) reported that clinical complexity factors underlying comorbidity were not a significant predictor of changes in disruptive behaviour as treatment responses. Similarly, van der Heijden et al. (2018) reported no association between sleep problems in children with ASD and comorbid anxiety and depression. Nonetheless, the findings from present study should be interpreted with caution as only eight out of 31 children had comorbid psychopathology. Of those eight children, only two children had comorbid anxiety disorder, three other children had ADHD. Hence, the present study might not have the power to investigate comorbidity and treatment responses.

Strengths of the current study

To our knowledge, this is one of the few studies that has used FBA to inform the development of sleep treatment in children with ASD (Jin et al., 2013; McLay et al., 2017, 2019; Singh & Zimmerman, 2015; Spruyt & Curfs, 2015). Given the various factors contributed to the development and maintenance of sleep problems, comprehensive FBA-informed assessments could identify behavioural, psychological and environmental factors unique to the individual and allow for treatment to be designed and implemented specifically targeting these factors. Such treatments would be more effective because the choice of intervention is driven by the variables precipitating and maintaining the behaviour and these variables were specifically addressed (McLay et al., 2017).

The data used in the present study was systematically collected in a larger Sleep and Autism study which employed a multiple-baseline across participants design. As suggested by Blampied (2001), several strengths of such research design were as follows. First, multiple repeated observations were performed on the individual level across consecutive observational or experimental phases. Second, functional analyses were performed at the individual level taking into account the uniqueness and singularity of participants. Third, analysis of therapeutic change was conducted based on the time-series data collected from each participant. Finally, clinical

innovations could be rigorously investigated and replications performed on multiple-baseline across participants to establish effectiveness and to map out the domain of generalisation (Blampied, 2001).

The results also add to the limited body of literature documenting the process of using individualized FBA informed interventions to treat a range of common sleep problems in children with ASD. In addition, this is one of the few studies that assessed the stability of treatment effects at both 4-6 weeks (i.e. short-term) and 10-12 weeks (i.e. long-term) post treatment. Most other studies have a much shorter follow-up period or limited follow-up (Christodulu & Durand, 2004; Delemere & Dounavi, 2018; Johnson et al., 2013; Kirkpatrick et al., 2019; Malow et al., 2014; Reed et al., 2009; Scantlebury, et al., 2018; Turner & Johnson, 2012) and long-term follow-up data would naturally be more convincing than post treatment data (Kazdin & Whitley, 2006).

Instead of relying solely on sleep diaries provided by parents or caregivers, the use of videosomnography in the present study allowed for verification of the reliability of the sleep diaries and at the same time gathering IOA data which further enhanced the reliability of observations. Videosomnography was found to be especially valuable in the present study since it was well tolerated by children with ASD and the data could be collected at home providing important information that might go undetected or falsely detected as awake by actigraphy (e.g., the child was awake but stayed sedentary while watching tablets or laptops; large amount of motor activity during periods of active rapid eye movement sleep (Hodge, Parnell, Hoffmann, & Sweeney, 2012; Moore, Evans, Hanvey, & Johnson, 2017; Sitnick, Goodlin-Jones, & Anders, 2008). Furthermore, the use of videosomnography also fulfilled the call for future research to use objective measure such as videotaping to collect IOA data (Delemere & Dounavi, 2018).

The involvement of parents as the primary agent in implementing treatments in the home environment is another strength of the present study contributing to the increasing body of literature that involves parental agency. The present results confirmed the effectiveness of the parent as an agent of change, consistent with past studies that utilized parents to implement treatment for sleep

disturbances (Austin et al., 2013; Malow et al., 2014; Meltzer & Mindell, 2014; Moon, Corkum, & Smith, 2010; Moss et al., 2014; Reed et al., 2009; Sanberg et al., 2018). Furthermore, the present study extends the practice of increasing parental agency and utilizing parent-derived sleep goals with increasing parental involvement using the Guided Participation Model (Sanders & Burke, 2014) which allows the development of sleep goals and treatment procedures in consultation and collaboration with parents (McLay et al., 2017). The use of the Guided Participation Model in the present study also helps to address possible barriers to treatment. Kazdin & Whitley (2006) suggested that barriers to treatment emerged in the context of parent contact with the treatment service, clinician, and intervention. Concern with the therapeutic relationship could be one barrier to treatment. By employing the Guided Participation Model, parents or caregivers were encouraged to actively participate in the intervention and decision-making. At the same time, clinicians aimed to establish and maintain an effective collaborative relationship with parents or caregivers in identifying treatment goals, in ensuring that parent goals are realistic and linked to appropriate strategies, and in empowering parents or caregivers to persist with change. Furthermore, clinicians were reminded to be alert to any signs of resistance, to address such resistance directly by exploring parents' perspectives, to take a non-judgmental stance that validated parents experience and to assist parents in exploring the advantages and disadvantages of making change (Sanders & Burke, 2014).

Effective behavioural treatments for sleep problems in children with ASD that can be reliably implemented within the home environment are limited (Sanberg et al., 2018). The present study demonstrated the effectiveness of multi-component and FBA informed treatments as implemented by parents or caregivers in a home setting. Being able to implement treatments at home is important since children with ASD might be averse to novel environments and a familiar environment might enhance the treatment responses (Moon et al., 2010; Moore et al., 2017; Sanberg et al., 2018).

Finally, a strength of the present study is that it addressed the concerns among clinicians regarding the generalizability of findings from controlled trials to clinical practice given the

possibility of differences between research samples and patients seen in clinics. Patients seen in clinics are more complex and diverse and no exclusion or inclusion can be applied whereas inclusion and exclusion criteria for study participants is a standard and required practice for high-quality research protocols (Patino & Ferreira, 2018). As mentioned above, children in present study were all from the larger Sleep and Autism study which employed single-case research design. The advantage of single-case research design allows for investigation to address the complexity presented at the individual level. To that end, children in the present study were as complex as patients seen in clinical practice given that all the children had a formal diagnosis of ASD and were either self-referral or referral by paediatricians or ASD case manager who was providing services to improve children's communication and behaviour to the families with children with ASD.

Limitations of the current study

The main limitation of this study is that only a single measure is used to capture each of the complexity variables (e.g., ASD symptom severity was measured using only the GARS; communication was measured using only the VABS communication sub-domain scores; and behaviour problems were measured using only the CBCL). Given that there were many aspects within each dimensions of complexity, using multiple measures would allow for a more comprehensive understanding of each child's functioning in a particular area (e.g., Anderson et al., 2009; Ben-Itzhak & Zachor, 2007; Sallows & Graupner, 2005; Kazdin & Whitley, 2006; Magiati et al., 2007; Perry et al., 2011; Remington et al., 2007). The second limitation of this study is the use of the SPS scoring system. The SPS scoring system, whilst derived from SBS and NSF Guidelines is not a validated measure. Hence, no psychometric testing had been conducted to assess the reliability (i.e., the ability to produce consistent results on sleep problem severity), validity (i.e., the ability to produce true results of sleep problem severity) and sensitivity (i.e., the probability of correctly identifying children with sleep problems) of the measure. Hence, further evaluation on the criterion validity (i.e., a degree of relationship between SPS score and CSHQ score) and construct validity (i.e., extent to which SPS assesses sleep problem severity and is associated with evidence

from objective measures such as videosomnography or polysomnography that measure sleep problems) would be desirable. Furthermore, SPS scores were only calculated based on set time periods (e.g., the final seven days of baseline). As a result, it may not reflect the complete picture of sleep problem severity during a particular phase, in its entirety. The third limitation of this study was the lack of comparison group given that all participants responded to treatment. Further classification of the group into responders and non-responders for comparison purposes were not possible. Future between-group comparison would be possible to better identify the characteristics of responders versus non-responders (Yoder & Compton, 2004). In contrast to Kazdin and Whitley's (2006) study which included socioeconomic disadvantage and parent and family functioning as two of the domains of case complexity, the present study did not investigate these factors. Kazdin and Whitley (2006) reported that both socioeconomic disadvantage and parent and family functioning were unrelated to treatment outcome. Nevertheless, given that there were many sources of complexity in relation to families, there was a need to investigate the family complexity factors. A separate and concurrent study on family complexity factors is underway using the same sample of children as the present study. It is also important to note that there was some missing data at baseline, short-term and long-term follow-up. However, due to the small sample size, no approach was taken to replace missing values. Hence, the present results should be preliminary and further replication would be needed. Finally, as cautioned by Vivanti et al. (2014), instead of actual predictors of outcomes, the factors proposed in the present study might only be the variables associated with change (i.e., mediators or moderators of treatment response) since not all the observed change can be attributed to treatment (Yoder & Compton, 2004). Given that there is no standard way to measure case complexity (Kazdin & Whitley, 2006), there is a possibility that failure to consider and to control factors other than treatment that might contribute to change and failure to include key dimensions of complexity would prevent the identification of correlates of changes that might truly predict the treatment responses (Kazdin & Whitley, 2006; Vivanti et al., 2014).

Recommendations for future research

On the basis of the aforementioned limitations, a number of recommendations have been made for future research. A key consideration is to use a greater variety of measures to capture complexity which would enable one to develop a more comprehensive understanding of the dimensions of complexity. For example, diagnostic instruments such as ADI-R, ADOS and CARS could be used to evaluate autism severity (e.g., Anderson et al., 2009; Ben-Itzhak & Zachor, 2007; Perry et al., 2011; Shui et al., 2018) and measures such as the Aberrant Behaviour Checklist (Kaat, Lecavalier, & Aman, 2014), Interview for Antisocial Behaviour (Kazdin & Esveldt-Dawson, 1986) and DBC could be used to measure challenging daytime behaviour (e.g., Kazdin & Whitley, 2006; Mazurek et al., 2019; Remington et al., 2007; Shui et al., 2018), and the Child Symptom Inventory – 4 (Sprafkin, Gadow, Salisbury, Schneider, & Loney, 2002) could be used to measure co-occurring psychiatric symptoms (e.g., Pellecchia et al., 2016).

Future research should attempt to objectively measure children's daytime behaviour. Using observational recordings to measure the frequency and duration of challenging daytime behaviour would be one example. Together with the use of sleep diaries to capture information about daytime naps or other information such as events at school, these combinations of objective and subjective measures would provide a better measure of children's daytime behaviour problems.

Further replications of key findings should be conducted and further investigation into the relationship between daytime behaviour and response to treatment is important. A recent pilot study had reported improvements in daytime behaviour (i.e., inattention, hyperactivity/noncompliance, repetitive behaviours and internalizing behaviours) following a brief behavioural intervention for insomnia in adolescents with ASD (Loring et al., 2018).

Clinical implications

The present study demonstrated the effectiveness of parent-implemented, function-based interventions to treat sleep problems in children with autism. Given that the latent variable comprising behaviour and sleep problems was a significant predictor of responses to treatment, an objective measure of daytime behaviour such as video recording will need to be used together with the parent- or caregiver-report CBCL score to prevent any bias associated with subjective measures. In terms of treatment, additional components to address behavioural problems will need to be formulated and included in the behavioural treatment package so that both behaviour and sleep problems can be addressed appropriately. Finally, the importance of a collaborative process in which decision making was driven by the child's parents or caregivers should not be underestimated to ensure success in the implementation and adherence to treatment plan.

Conclusion

As demonstrated in the present study, reduction in sleep problem severity could be achieved through the use of a multi-component, parent-implemented, FBA informed treatments. In addition, the present study suggested that the severity of sleep and behaviour problems was the strongest predictor of response to treatment. Comorbidity, autism symptoms severity, medications and communications did not seem to predict response to treatment. Overall, the study added to the current literature in terms of identifying possible predictors of treatment outcomes and effectiveness of functions-based interventions to treat sleep problems in children with ASD. A multimodal measurement of improvements observed that include objective measures such as videosomnography and subjective measures in addition to a greater variety of measures to capture complexity dimensions were recommended for future research to further improve the validity and utility of the SPS score. Finally, even if the treatment seemed to be less effective with more complex cases (i.e. those with more behaviour or sleep problems), that would not mean the treatment was ineffective, as shown by the small to medium effect size when categorized using behaviour problems and medium to large effect size when categorized using sleep problems. The present study suggested

that both more complex and less complex cases could respond to treatment and case complexity was not a deterrent to responses to treatment.

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Appendices

Appendix A. Child Information Sheet



An investigation into the efficacy of treatments for sleep disturbance in children with autism

Children's Information Sheet

Hello. My name is XX and I am a XX student at the University of Canterbury. I am doing a project about how to help children to sleep better and I would like for you to help me with this.

I am going to be talking to you and your parent/s about ways to help you to sleep better. This means that I might be Skyping you, coming to your house, or your parent/s will be coming to see me at the University.

There will be a video camera in your bedroom sometimes. This will help me to understand what you do when you are awake and asleep. Only your parents and other people working on this project will be able to see this video. We may ask you to wear an actigraph. An actigraph is worn on your wrist like a watch and it tells us when you are asleep and when you are awake.

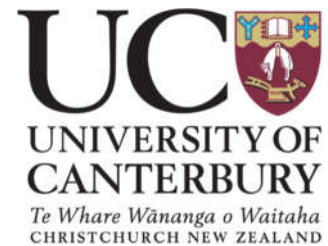
If you do not want to be a part of this project, you can tell me or your parents and you won't need to be a part of it anymore.

If you have any questions you can ask me or your parents whenever you like.

Now we need to decide if you would like to do this. If you do want to be a part of my project then you can say "yes". If you do not want to be a part of this project then you can say "no" and no one will mind.

If you say yes, you or one of your parents can sign the form for you.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email human-ethics@canterbury.ac.nz



“An investigation into the efficacy of treatments for sleep disturbance in children with autism”

Children’s Consent Form

My name is _____.

- ☐ XXX has told me about the work that she is going to be doing with me and my parent/s.
- ☐ XXX told me that she is going to be working with me and my parent/s to help me to learn to sleep better.
- ☐ While XXX does this she will be asking my parents about my sleep each night and there will be a video camera in my room on some nights that is recording my sleep.
- ☐ I know that if at any time I want to stop being a part of this project then XXX will stop recording data and this will be destroyed.
- ☐ If I want XX to stop video recording my sleep then the camera will be taken out of my room and that will be fine. If I want any video footage to be deleted, I can tell XXX or my parents.
- ☐ I was told that my parents/caregiver may sign this form for me and I think that is OK.
- ☐ I would like a summary of the results of this project.

Child’s name: _____

Date: _____

Signature: _____

If this form is signed on behalf of your child please acknowledge, by signing this form, that your child was verbally informed of the investigation and what it will involve and that they were unable to provide verbal or written consent that they would like to be a part of this research.

Parent/caregiver: _____

Date: _____

Signature: _____

Please return this form to XXX.

An investigation into the effectiveness of treatments for sleep disturbance in children with autism or features of autism

Information for Parents/Caregivers

This research has been assessed and approved by the University of Canterbury Human Ethics Committee (HEC 2018/47).

Dear Parent/ Caregiver,

We are a group of researchers at the University of Canterbury. Dr Laurie McLay is a Senior Lecturer in the School of Health Sciences at the University of Canterbury. Laurie has many years experience in working with children and young people with developmental disabilities and their families. Associate Professor Karyn France has lectured here for many years, has conducted research into the treatment of paediatric sleep disturbance and is a registered clinical psychologist with considerable clinical experience in this area. Professor Neville Blampied has a similar history of teaching and research. A number of Masters and PhD students and Child and Family Intern psychologists or registered psychologist also work on this project.

We would like you and your child with autism to consider participating in this research study. The primary purpose of this study is to investigate the effectiveness of treatments for sleep disturbance in children with autism. Treatment can include a range of strategies, including both non-traditional approaches (such as white noise) and behavioural interventions. These approaches have been designed to minimise stress as much as possible for the parents and children using them. We are also interested in parents' and children's experiences in using the treatments and any changes to their lives, or their child's lives, which result.

As a part of this study we would also like to investigate the experiences of parents in implementing treatments for sleep disturbance, those treatments that they consider to be most acceptable, and the impact of successful treatment of sleep problems on parent and child wellbeing and quality of life. In order to do this we will ask you to complete some questionnaires about you and your child's well-being and behaviour at the commencement and conclusion of treatment. We will also ask your perspective on the treatment that was provided. We will do this either during visits to your home, Skype interviews, or in a clinic at the University of Canterbury.

If you agree to allow your child to be a part of this study, we will meet with you, or Skype you, to discuss your child's sleep behaviour and find out more about him/her and your family. This initial

meeting will last for approximately 1-1 ½ hours. We will then ask you to complete sleep diaries in which you will record further information about your child's sleep patterns. Sleep diaries will be recorded each day throughout all phases of the study as this will allow us to monitor the effectiveness of the treatment approach. The sleep diaries will take you up to five minutes to complete each night. You will also be asked to complete commonly used questionnaires in order to obtain information about your child's sleep behaviour and the effects of treatment. It will take approximately 15 minutes to complete each questionnaire. When we have established an understanding of your child's sleep behaviour, we will work with you to develop sleep-related goals for your child. This will involve a second treatment planning session which will last 1-1 ½ hours.

To help us gather further information about your child's sleep patterns we will bring or send a video camera to your home for some nights over the course of the programme, which is capable of recording all night sleep. In addition we may ask you, if possible, to use an actigraph with your child. This watch-like device records the movements associated with sleep and can be worn on the wrist or ankle, or secured into a pocket on your child's pyjamas. This may offer an alternative to video cameras when appropriate. These methods will allow us to measure sleep behaviour at times when an adult is not present. We will demonstrate and explain how to use each of these methods for gathering information.

When information about your child's sleep behaviour has been gathered, treatment will commence. You will be offered a choice of treatment options, which you will then implement with the support of the research team. If you are dissatisfied with the treatment approach or the degree of progress that is being made then you will be offered a choice of another treatment option. We will provide you with all of the necessary information about each treatment approach and we will maintain regular contact with you during treatment. It is anticipated that your involvement in the study will occur over the course of a few months, but will depend on the rate of your child's progress as well as your satisfaction with the progress.

For the purpose of this project, myself (insert name), a psychologist/intern psychologist/Masters/PhD student will be working closely with you to conduct the necessary assessments and formulate interventions. XX, a research assistant/Masters/PhD student who also works as a part of our sleep team, may look at some of the information that we collect, such as video recordings and actigraph data.

Your child will be assigned a code name to ensure anonymity and anything that you or your child says or does will be kept confidential. The results of the study may be submitted for publication to national or international journals and may also be presented at conferences. No identification of the child or family will be possible from any report, publication or presentation.

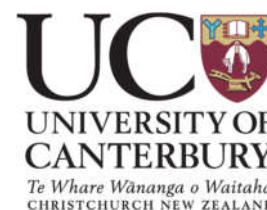
If you want to withdraw from the project before completion, you can do this at any time without penalty or repercussions.

Should you require any additional information about the study or if you would like to access the study findings you are able to do so at any stage. The data which is produced from the research will be kept in a locked cabinet at the University of Canterbury for a minimum of ten years.

If you agree for your child to take part in the research, please sign the consent form that is attached.

If you have any complaints you may contact the Chair of the University of Canterbury Ethics Committee. The contact details are given below.

If you have any questions about this project please feel free to contact Dr Laurie McLay: Phone (03) 369-3522 or, email: laurie.mclay@canterbury.ac.nz



An investigation into the effectiveness of treatments for sleep disturbance in children with autism

CONSENT FORM FOR PARENTS/ CAREGIVERS

This research has been assessed and approved by the University of Canterbury, Human Ethics Committee (HEC 2018/47).

- ☐ I wish to participate in the project, “An investigation into the efficacy of treatments for sleep disturbance in children with autism”
- ☐ I have read and been given a full explanation of this project and have had the opportunity to ask questions.
- ☐ I understand what will be required of myself and my child/the child in my care during this project.
- ☐ I understand that the investigators do not foresee any potential risks to me or my child as a result of participating in this study. However, if the intervention results in an increase in family stress, the staff working with us will provide support.
- ☐ I understand that all information about my family will be treated as confidential unless there is concern about anyone’s safety. In this case my clinician will need to speak to someone else to ensure the safety risk is removed. No findings that could identify me or my child will be published
- ☐ I understand that the findings of this study may be published in a research journal or at a conference and that the anonymity of my child and I will be maintained
- ☐ I understand that participation in this project is voluntary and that I can withdraw my child or he/she can withdraw from the project at any time without repercussions. I can also withdraw any data that has been collected at any time prior to the publication of that data
- ☐ I understand that all research data that is collected will be securely stored at the University of Canterbury for a minimum of ten years
- ☐ I understand that I am able to request a copy of the results of this research, should I wish to do so, and that these results will be provided for me

- ☐ I allow video-taping of my child's sleep behaviour to be completed by the researcher and understand that this videotape will be used for data gathering purposes only. I also understand that I have the right to request that video footage is destroyed at any stage.
- ☐ I consent to others, listed below, being involved in the implementation of the intervention

Name: _____

Date: _____

Signature: _____

Others I consent to implementing intervention:

Name: _____

Name: _____

Name: _____

- ☐ I would like a summary of the results of this project.

Please return this form to XXX.


Appendix E. Sleep Diary Template

	Date:	Monday:	Tuesday:	Wednesday:	Thursday:	Friday:	Saturday:	Sunday:
Daytime sleep	Setting (where fell asleep)							
	Time asleep							
	Time awake							
Night-time sleep	Setting (where fell asleep)							
	Time put to bed							
	Frequency of Curtain calls*							
	Curtain calls after put to bed (Describe each)							
	Your responses to each curtain call (Describe each)							
	Best estimate of time asleep							

Sleep Diary

Child's Name:

		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
1 st Night time awakening	Time & Duration of awakening	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
<hr/>								
		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
2 nd Night time awakening	Time & Duration of awakening	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
<hr/>								
		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday

3 rd Night time awakening	Time & Duration of awakening	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins	_____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
								
Time awake in the morning								

- Curtain calls: Any behaviour such as leaving the bed (or bedroom) or calling parents into the room, between the time of being put to bed and falling asleep

Notes:

**An Investigation into the Efficacy of Treatments for Sleep Disturbance in
Children with Autism**

AUDIOVISUAL RECORDING CONSENT FORM

You have been given this form because the researchers have asked your permission to take audiovisual recordings of your child's sleep behavior.

Please read the statements below, which explain the purpose of audiovisual recording and how your privacy will be protected:

- The purpose of recording is to gather data for the research project
- Audiovisual recording will only be done with your knowledge and consent
- You can withdraw your consent to audiovisual recording at any time, without having to provide a reason for changing your mind
- The audiovisual file will only be seen by the researchers
- The audiovisual recording will be securely stored at the University of Canterbury for a minimum of ten years

I hereby consent to audiovisual recordings being made on the above conditions.

Signed: _____

Date: _____